

Intussusception due to rectal adenocarcinoma in a young adult: A case report

Ryo Inada, Takeshi Nagasaka, Toshiaki Toshima, Yoshiko Mori, Yoshitaka Kondo, Hiroyuki Kishimoto, Toshiyoshi Fujiwara

Ryo Inada, Takeshi Nagasaka, Toshiaki Toshima, Yoshiko Mori, Yoshitaka Kondo, Hiroyuki Kishimoto, Toshiyoshi Fujiwara, Department of Gastroenterological Surgery, Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, Okayama 700-8558, Japan

Author contributions: Inada R wrote this report; Nagasaka T and Mori Y analyzed microsatellite status; Toshima T, Kondo Y and Kishimoto H collected the patient's clinical data; Nagasaka T and Fujiwara T edited this report.

Correspondence to: Takeshi Nagasaka, MD, PhD, Department of Gastroenterological Surgery, Okayama University Graduate School of Medicine, Dentistry and Pharmaceutical Sciences, 2-5-1 Shikata-cho, Kita-ku, Okayama 700-8558, Japan. takeshin@cc.okayama-u.ac.jp

Telephone: +81-86-235-7257 Fax: +81-86-221-8775

Received: April 9, 2014 Revised: May 22, 2014

Accepted: June 14, 2014

Published online: September 21, 2014

Abstract

An intussusception due to colonic adenocarcinoma has sometimes been reported. However, to the best of our knowledge, reports of intussusception due to rectal adenocarcinoma are extremely rare. In this report, the case of a young man with rectal adenocarcinoma causing intussusception is described. A 24-year-old man visited a hospital complaining of abdominal pain, and an upper rectal cancer was diagnosed by colonoscopy. Computed tomography showed intussusception caused by a large tumor in the pelvis and absence of distant metastases. Locally advanced rectal cancer causing intussusception was diagnosed, and a low anterior resection was performed. Intraoperatively, repair of the invagination could not be accomplished easily; therefore, the repair was abandoned. Instead, the tumor was removed *en bloc* to avoid dissemination of the cancer. Histopathologically, the tumor was diagnosed as a poorly differentiated adenocarcinoma, pStage II A. The patient has no evidence of recurrence at 10 mo after the operation.

© 2014 Baishideng Publishing Group Inc. All rights reserved.

Key words: Adult intussusception; *En bloc* resection; Low anterior resection; Rectal adenocarcinoma; Young cancer

Core tip: Intussusception is uncommon in adults compared with children, and colonic tumors have sometimes been reported as causes of adult intussusception. On the other hand, there have been only a few reports of intussusception due to rectal adenocarcinoma in the English literature. In this report, an extremely rare case of sporadic rectal adenocarcinoma causing intussusception in a young man who underwent curative resection and has survived is described.

Inada R, Nagasaka T, Toshima T, Mori Y, Kondo Y, Kishimoto H, Fujiwara T. Intussusception due to rectal adenocarcinoma in a young adult: A case report. *World J Gastroenterol* 2014; 20(35): 12678-12681 Available from: URL: <http://www.wjgnet.com/1007-9327/full/v20/i35/12678.htm> DOI: <http://dx.doi.org/10.3748/wjg.v20.i35.12678>

INTRODUCTION

Intussusception is a common disease and one of the leading causes of intestinal obstruction and acute abdominal emergencies in children^[1]. On the other hand, adult intussusception is unusual, accounting for only 5% of all cases of intussusception and 1%-5% of all cases of bowel obstruction^[2-4]. The cause of intussusception differs between the pediatric and adult populations. In adults, a demonstrable etiology is present in about 70%-90% of all cases with intussusception, whereas about 90% of pediatric cases of intussusception are idiopathic^[1,5-8]. Intraluminal lesions form leading edges of most cases of adult intussusception. Malignant tumor

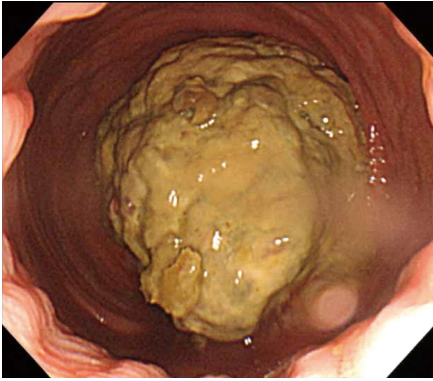


Figure 1 Colonoscopy showing a huge tumor at the upper rectum. The tumor, measuring 100 mm in diameter, does not show invagination on colonoscopy.



Figure 2 Magnetic resonance imaging showing intussusception caused by a tumor in the pelvis. The upper rectal tumor (asterisk) causes intussusception (arrowheads) as a leading edge.

is often found as the etiology of adult intussusception, and colonic adenocarcinomas are sometimes reported as causes of intussusception^[5,6,9-12]. However, there appear to be only a few reports of intussusception due to rectal adenocarcinoma in the English literature^[13]. Therefore, an extremely rare case of a young man with sporadic rectal adenocarcinoma causing intussusception who underwent curative resection and has survived is described.

CASE REPORT

A 24-year-old man was referred for management of rectal cancer. Initially, he had visited another hospital with a complaint of abdominal pain and tenesmus, and an upper rectal tumor measuring 100 mm in diameter, without invagination, was diagnosed by colonoscopy (Figure 1). He had a family history of gastrointestinal cancer; his maternal uncle and grandmother had died of colorectal cancer, and his paternal grandfather had died of gastric cancer. At the time of his transfer to our hospital, laboratory tests were within the normal range, including a carcino-embryonic antigen level of 0.83 ng/mL and a carbohydrate antigen 19-9 (CA19-9) level of 9.2 U/mL. Computed tomography (CT) and magnetic resonance imaging showed intussusception caused by a large tumor in the pelvis and absence of distant metastases (Figure 2). Locally advanced upper rectal cancer causing intussusception was diagnosed, and a low anterior resection with



Figure 3 Specimen resected in *en bloc* fashion. The tumor is located at the upper rectum, and the margin is negative for cancer. Pathological staging is pStage II A (T3, N0, M0).

complete removal of all regional lymph nodes and total mesorectal excision was performed. Intraoperatively, the invagination could not be repaired easily; therefore, the repair was abandoned. Instead, the tumor was removed *en bloc* to avoid dissemination of the cancer. After the proctectomy, coloproctostomy with a temporary diverting ileostomy was performed. Histopathologically, the surgical margin was negative for cancer, and based on the classification of the seventh edition of the Union for International Cancer Control (UICC), the tumor was diagnosed as a poorly differentiated adenocarcinoma, pStage II A (T3, N0, M0) (Figures 3 and 4)^[14]. To exclude Lynch syndrome, microsatellite status and immunohistochemistry of four deoxyribonucleic acid (DNA) mismatch repair proteins, including mutL homolog 1, postmeiotic segregation increased 2, mutS homologue 2 and mutS homologue 6, were checked; the tumor was microsatellite stable and retained expression of all mismatch repair proteins on immunohistochemistry.

The patient underwent adjuvant XELOX (capecitabine plus oxaliplatin) for 6 mo, and 9 mo after the low anterior resection, he was alive without evidence of recurrence.

DISCUSSION

Adult intussusception is uncommon and has some different characteristics from pediatric intussusception. Pediatric patients with intussusception often show acute symptoms of abdominal pain, bloody stools and a palpable abdominal mass, the so-called classic presentation triad^[1]. On the other hand, adult patients with intussusception rarely show the complete triad^[9]. They have subacute or chronic symptoms caused by intestinal obstruction, including abdominal pain, nausea and vomiting^[5,9-12]. According to some previous retrospective institutional reviews of 20-60 cases of adult intussusception treated surgically^[5,9-12], colonic intussusception is minor (15.0%-35.0%) compared with enteric intussusception. Most cases of adult intussusception are caused by intraluminal lesions, and malignant tumors are responsible for 22.2%-46.6% of adult intussusception cases^[5,9-12]. With regard to colonic adenocarci-

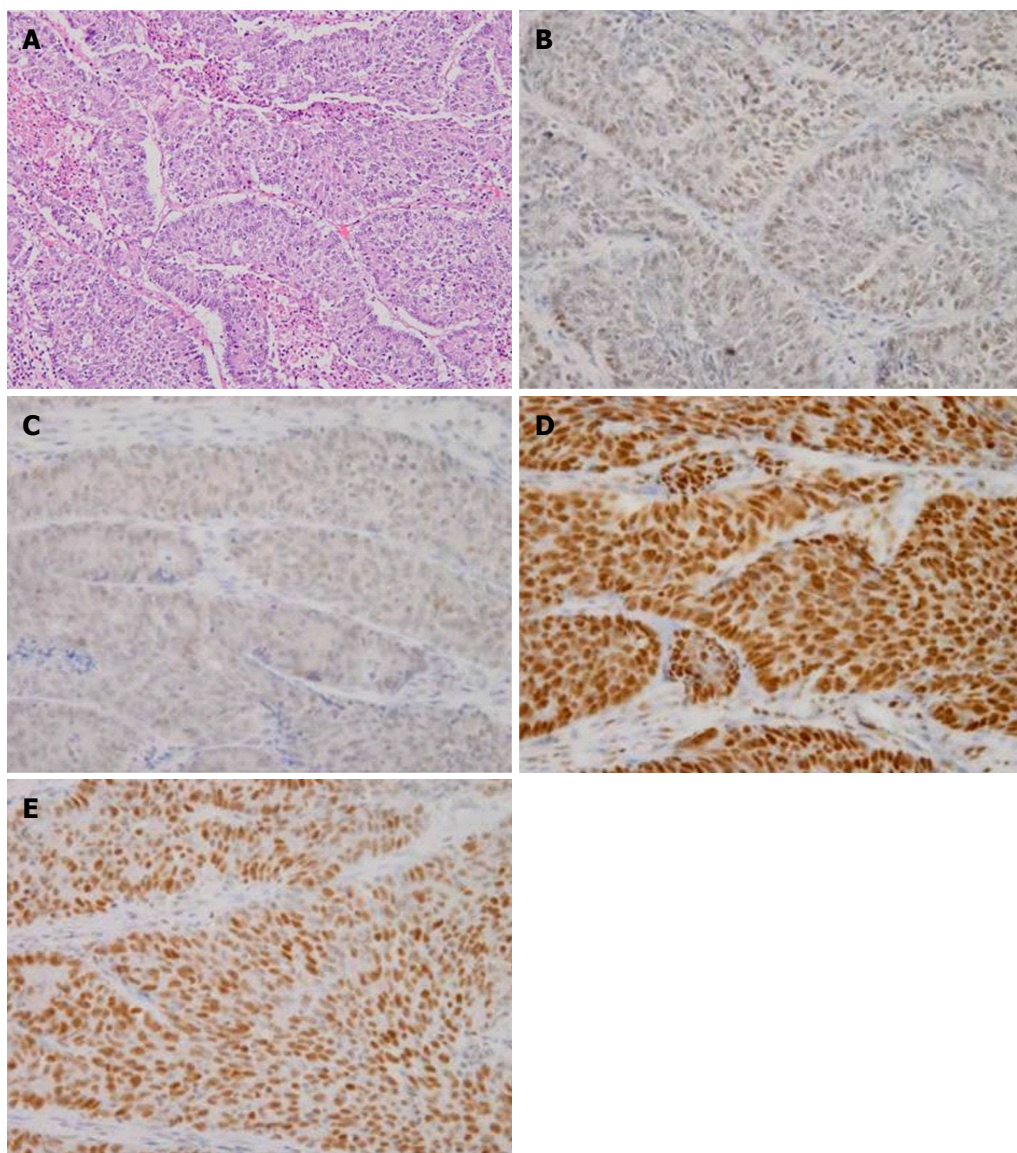


Figure 4 Microscopic findings and immunohistochemistry for deoxyribonucleic acid mismatch repair proteins ($\times 200$). The tumor is a poorly differentiated adenocarcinoma. Immunohistochemical expression of mutL homolog 1 (MLH1), postmeiotic segregation increased 2 (PMS2), mutS homolog 2 (MSH2) and mutS homolog 6 (MSH6) is retained. HE: Hematoxylin and eosin stain. A: HE; B: MLH1, C: PMS2; D: MSH2, E: MSH6.

noma, it accounted for 33.3%-80.0% of cases of colonic intussusception and 7.3%-25% of all adult intussusception cases^[5,9-12]. A literature search was conducted, and it identified only a few English manuscripts, including case reports, about rectal intussusception caused by rectal adenocarcinoma^[13]. In this case, a preoperative diagnosis of intussusception was made by computed tomography, but colonoscopy did not show the intussusception. It is sometimes difficult to diagnose adult intussusception preoperatively; the rate of preoperative diagnosis is reported to range from 32% to 70%^[5,9-12]. The accuracy of computed tomography for preoperative diagnosis of adult intussusception is better than that of other examinations, including abdominal ultrasonography, barium enema study, and colonoscopy^[5,9-12].

In the present case, the patient was diagnosed as having rectal cancer at a young age. Young patients with colorectal cancers are sometimes encountered, and they more frequently have Lynch syndrome, which is caused

by germline mutations of DNA mismatch repair genes^[15]. Microsatellite status and immunohistochemistry for DNA mismatch repair proteins were examined for the present tumor, but the patient did not have Lynch syndrome.

In conclusion, an extremely rare case of a young man with a sporadic rectal adenocarcinoma causing intussusception was reported.

ACKNOWLEDGMENTS

The authors would like to thank Dr Hiroko Toda and Hiroyuki Yanai for help in making the pathological diagnosis.

COMMENTS

Case characteristics

A 24-year-old man with a family history of gastroenterological cancers.

Clinical diagnosis

Abdominal pain and tenesmus.

Differential diagnosis

Infectious enteritis and inflammatory bowel disease.

Laboratory diagnosis

Carcino-embryonic antigen 0.83 ng/mL, carbohydrate antigen 19-9 9.2 U/mL and other laboratory data were within normal limits.

Imaging diagnosis

Colonoscopy showed an upper rectal tumor, and computed tomography showed intussusception caused by a large tumor in the pelvis and absence of distant metastases.

Pathological diagnosis

The tumor was diagnosed as a poorly differentiated adenocarcinoma, pStage II A.

Treatment

The patient underwent a low anterior resection with complete removal of all regional lymph nodes and total mesorectal excision and adjuvant XELOX (capecitabine plus oxaliplatin).

Related reports

Reports of intussusception due to rectal adenocarcinoma are extremely rare.

Term explanation

Microsatellite status and immunohistochemistry for deoxyribonucleic acid mismatch repair proteins used for diagnosis of Lynch syndrome and sporadic microsatellite instability cancers.

Experiences and lessons

If the invagination cannot be repaired easily, we should abandon the repair and remove the tumor *en bloc* to avoid dissemination of the cancer.

Peer review

The authors described an extremely rare case of a young man with a sporadic rectal adenocarcinoma causing intussusception in this manuscript.

REFERENCES

- Jiang J, Jiang B, Parashar U, Nguyen T, Bines J, Patel MM. Childhood intussusception: a literature review. *PLoS One* 2013; **8**: e68482 [PMID: 23894308 DOI: 10.1371/journal.pone.0068482]
- Laws HL, Aldrete JS. Small-bowel obstruction: a review of 465 cases. *South Med J* 1976; **69**: 733-734 [PMID: 935902 DOI: 10.1097/00007611-197606000-00021]
- Stewardson RH, Bombeck CT, Nyhus LM. Critical operative management of small bowel obstruction. *Ann Surg* 1978; **187**: 189-193 [PMID: 629620 DOI: 10.1097/00000658-197802000-00017]
- Donhauser JL, Kelly EC. Intussusception in the adult. *Am J Surg* 1950; **79**: 673-677 [PMID: 15410943 DOI: 10.1016/0002-9610(50)90333-3]
- Azar T, Berger DL. Adult intussusception. *Ann Surg* 1997; **226**: 134-138 [PMID: 9296505]
- Sanders GB, Hagan WH, Kinnaird DW. Adult intussusception and carcinoma of the colon. *Ann Surg* 1958; **147**: 796-804 [PMID: 13534248 DOI: 10.1097/00000658-199708000-00003]
- Nagorney DM, Sarr MG, McIlrath DC. Surgical management of intussusception in the adult. *Ann Surg* 1981; **193**: 230-236 [PMID: 7469558 DOI: 10.1097/00000658-198102000-00019]
- Begos DG, Sandor A, Modlin IM. The diagnosis and management of adult intussusception. *Am J Surg* 1997; **173**: 88-94 [PMID: 9074370 DOI: 10.1016/S0002-9610(96)00419-9]
- Wang N, Cui XY, Liu Y, Long J, Xu YH, Guo RX, Guo KJ. Adult intussusception: a retrospective review of 41 cases. *World J Gastroenterol* 2009; **15**: 3303-3308 [PMID: 19598308 DOI: 10.3748/wjg.15.3303]
- Yakan S, Caliskan C, Makay O, Denecli AG, Korkut MA. Intussusception in adults: clinical characteristics, diagnosis and operative strategies. *World J Gastroenterol* 2009; **15**: 1985-1989 [PMID: 19399931 DOI: 10.3748/wjg.15.1985]
- Eisen LK, Cunningham JD, Aufses AH. Intussusception in adults: institutional review. *J Am Coll Surg* 1999; **188**: 390-395 [PMID: 10195723 DOI: 10.1016/S1072-7515(98)00331-7]
- Goh BK, Quah HM, Chow PK, Tan KY, Tay KH, Eu KW, Ooi LL, Wong WK. Predictive factors of malignancy in adults with intussusception. *World J Surg* 2006; **30**: 1300-1304 [PMID: 16773257 DOI: 10.1007/s00268-005-0491-1]
- Mussack T, Szeimies U. Sigmoidorectal intussusception caused by rectal carcinoma: multislice CT findings. *Abdom Imaging* 2002; **27**: 566-569 [PMID: 12173000 DOI: 10.1007/s00261-001-0105-9]
- Sobin LH, Gospodarowicz MK, Wittekind C. TNM Classification of Malignant Tumours, 7th Edition. UK, Chichester: Wiley-Blackwell, 2009
- Boland CR, Thibodeau SN, Hamilton SR, Sidransky D, Eshleman JR, Burt RW, Meltzer SJ, Rodriguez-Bigas MA, Fodde R, Ranzani GN, Srivastava S. A National Cancer Institute Workshop on Microsatellite Instability for cancer detection and familial predisposition: development of international criteria for the determination of microsatellite instability in colorectal cancer. *Cancer Res* 1998; **58**: 5248-5257 [PMID: 9823339]

P- Reviewer: Rekhi B

S- Editor: Nan J L- Editor: O'Neill M E- Editor: Du P





Published by **Baishideng Publishing Group Inc**

8226 Regency Drive, Pleasanton, CA 94588, USA

Telephone: +1-925-223-8242

Fax: +1-925-223-8243

E-mail: bpgoffice@wjgnet.com

Help Desk: <http://www.wjgnet.com/esps/helpdesk.aspx>

<http://www.wjgnet.com>



ISSN 1007-9327

