

Two case reports

Colorectal adenocarcinoma in children

Chang hoon Ahn, MD, Soon Chul Kim, MD, PhD*

Abstract

Rationale: Colorectal cancer in children is rare, with delayed diagnosis and advanced stage at presentation in high mortality. Early detection of colorectal cancer is, therefore, important for better prognosis.

Patient concerns: Thirteen-year-old boy presented with symptoms of melena, vomiting, and abdominal pain for 6 months. 18-year-old girl was hospitalized due to the symptoms of hematochezia, and persistent abdominal pain for 6 months. They have no chronic disease or familial history of malignancy.

Diagnoses: We encountered 2 teenagers diagnosed with colorectal cancer.

Interventions: Both patients had the same histological findings in postoperation colonic biopsy and underwent surgical resection.

Outcomes: The boy fully recovered with only surgery, but the girl died, despite receiving adjuvant chemotherapy for the advanced stage of cancer.

Lessons: We recommend early and active evaluation, including a pediatric colonoscopy, in a child with suspected malignancy.

Abbreviations: CA = cancer antigen, CEA = carcinoembryonic antigen, CT = computed tomography, DIC = disseminated intravascular coagulation.

Keywords: adenocarcinoma, children, colon cancer

1. Introduction

In the pediatric group aged ≤ 20 years, the incidence of colorectal cancer is rare compared with that in adults. Colorectal cancer can be fatal, but early diagnosis and improved management has resulted in reduced mortality since 1980s.^[1] Therefore, the cancer screening system in Korea has adopted routine colorectal endoscopy for individuals aged over 40 years since 1999.^[2] However, routine colonoscopy is not recommended in younger individuals, resulting in advanced stages of colorectal cancer at

presentation in patients younger than 20 years.^[3–5] Regardless of the low incidence of colorectal cancer in children, young patients with gastrointestinal symptoms of suspected colorectal cancer should be screened for it. The stage of colorectal cancer, like the histological type of cancer, is 1 of the most important factors determining the mortality in such patients. In this report, we describe 2 pediatric cases diagnosed with the colorectal adenocarcinoma, each with different course of the disease. With our brief case reviews, we emphasize the importance of early detection of colorectal cancer.

Written informed consent was obtained from the patients before the publication of this case report and accompanying images.

2. Case 1

A 13-year-old boy was hospitalized because of acute abdominal pain, melena, and vomiting. Mild abdominal pain was present for the preceding 6 months in the right lower quadrant. No weight loss or other specific symptoms were present. Before the onset of the symptoms, no signs of malignancy were present and he had not undergone a thorough abdominal examination till then. Additionally, he did not have any chronic diseases and had no family history of malignancy. Distended abdomen and decreased bowel sounds were observed during the physical examination. Diffuse direct and indirect tenderness was present throughout the abdomen. At presentation, melena was present; however, blood pressure and other vital signs were normal.

Plain radiography indicated a suspicious state of ileus; however, we could not see the exact mechanical obstruction. Physical examination and radiographic findings warranted further examination; hence, in view of the patient's age, we ordered abdominal computed tomography (CT) scan immediately. Abdominal CT scan confirmed the presence of ileocolic

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Department of Pediatrics, Chonbuk National University Medical School and Hospital, Research Institute of Clinical Medicine of Chonbuk National University - Biomedical Research Institute of Chonbuk National University Hospital, Jeonju, Korea.

* Correspondence: Soon Chul Kim, Department of Pediatrics, Chonbuk National University Hospital, 20 Geonji-ro, Deokjin-gu, Jeonju 54907, Korea (e-mail: kimsc@jbnu.ac.kr).

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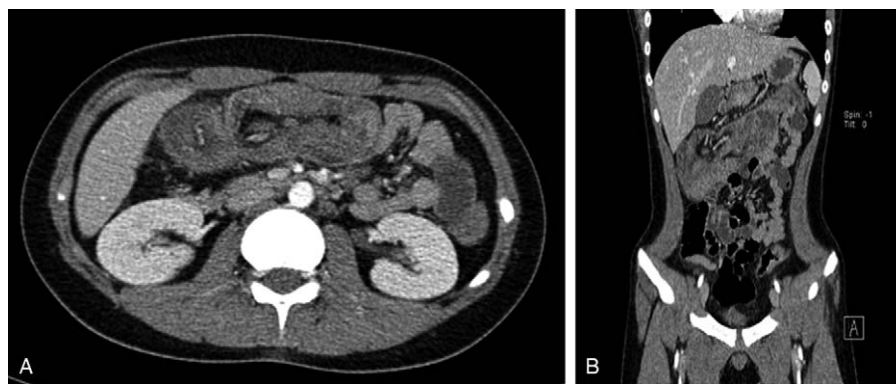


Figure 1. Abdomen and pelvic CT. (A) It shows inversion of distal ileum, mesenteric fat, and multiple mesenteric lymph node enlargement. (B) Coronal image shows same findings. CT=computed tomography.

intussusception with distal segmental ischemic changes (Fig. 1). Because the patient had visited our hospital after 24 hours of the onset of his symptoms with suspected ileocolic obstruction, we decided to intervene with emergency surgical resection. The initial laboratory results revealed anemia (hemoglobin = 7.1 g/dL) and he had ongoing melena; therefore, red blood cell transfusion was performed. Inflammatory markers such as white blood cell count, C-reactive protein level, and erythrocyte sedimentation rate were normal. Platelet count and disseminated intravascular coagulation (DIC) score were also within the normal range. These normal laboratory results and a lack of familial history made bleeding diathesis highly unlikely. Since levels of tumor markers like carcinoembryonic antigen (CEA) and cancer antigen (CA) 19-9 were within the normal range, we could not find any indication of malignancy before histological results in postoperative colonic biopsy were revealed.

Only segmental resection was planned initially, but juvenile polyposis was suspected during the surgery, and, therefore, right hemicolectomy was performed (Fig. 2). Histological results confirmed well-differentiated adenocarcinoma with no evidence of metastasis (Fig. 3). Finally, he was diagnosed with stage I colorectal cancer and additional treatment such as adjuvant chemotherapy was not necessary. Postoperatively, blood tests, radiology screening, and colonoscopy were performed annually.



Figure 2. The specimen that is eliminated from right colon. It shows multiple palpable soft polypoid mass on right ascending colon.

No signs of recurrent colorectal cancer were found during these tests.

3. Case 2

An 18-year-old girl presented with acute abdominal pain and hematochezia. She had experienced abdominal pain at least 3 times and lost over 10 kg weight during the past 6 months. She also did not have any chronic diseases or gastrointestinal problems before the onset of symptoms. No one in her family had colorectal or other malignancies. Physical examination revealed normal vital signs, decreased bowel sounds, severely distended abdomen, and direct tenderness in the right lower quadrant. Digital rectal examination revealed hematochezia. L-tube drainage did not show signs of upper gastrointestinal bleeding. Vital signs were normal.

Plain radiography revealed mechanical obstruction (Fig. 4A) and abdominal CT scan indicated adenocarcinoma of the descending colon, with peritoneal seeding (Fig. 4B). Owing to the severe abdominal distension, colonoscopy was performed to relieve the gas and to confirm the diagnosis. Colonoscopic

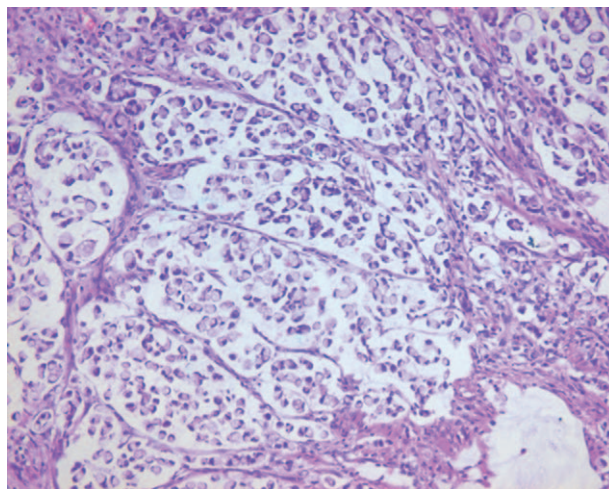


Figure 3. Histological finding. The cancer was confirmed with adenocarcinoma, well-differentiated type (H&E $\times 400$). The tumor invaded in the submucosa and regional lymph node, but metastasis was not found. H&E=hematoxylin and eosin.

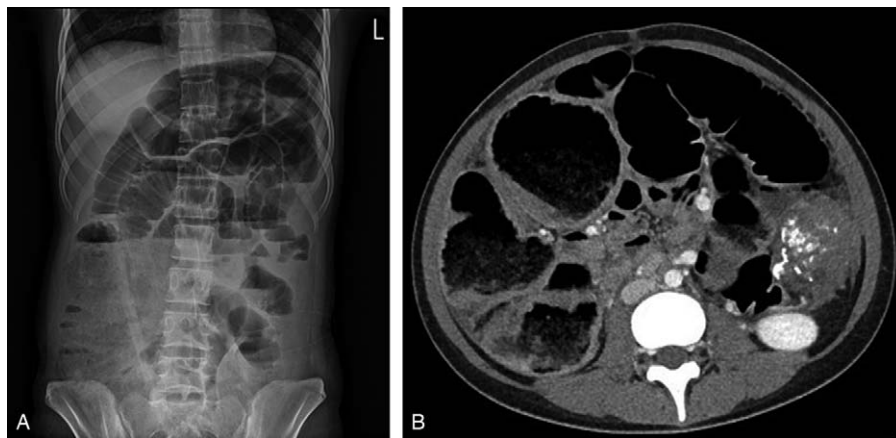


Figure 4. (A) Simple abdominal x-ray (B) abdominal CT. It shows paralytic ileus due to mechanical obstruction, suspicious of adenocarcinoma with peritoneal seeding. CT=computed tomography.

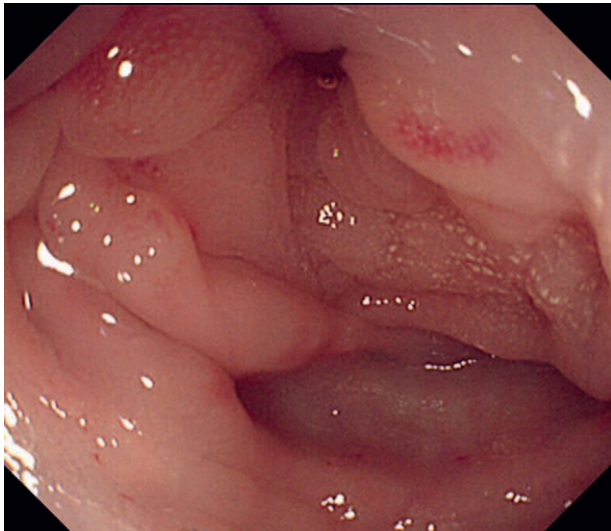


Figure 5. Colonoscopy. It shows severe edematous mucosa in right ascending colon and the scope could not pass because of obstruction.

findings included edematous mucosa with severe stenosis and obstruction of the descending colon that did not allow inserting the scope further (Fig. 5). Initial laboratory test results revealed mild anemia (hemoglobin = 11.5 g/dL), but white blood cell and platelet counts were normal. Activated partial thromboplastin time and DIC scores were within the normal range. These findings with a lack of familial history of hereditary bleeding disorders indicated low probability of a bleeding disorder. However, high levels of tumor marker CA 125 (43.90 U/mL) were found.

We decided to perform an emergency surgical resection and a subtotal colectomy (Fig. 6). Histological examination revealed poorly differentiated mucinous adenocarcinoma that had penetrated the surface of the visceral peritoneum (Fig. 7). Regional lymph node metastasis and distant metastasis were also found by positron emission tomography/CT and bone scans after the surgery. Finally, she was diagnosed with stage IV colorectal cancer and adjuvant chemotherapy (Avastin, FOLFIRI) was administered. During the course of chemotherapy, she developed severe abdominal pain and recurrent ileus. However, her condition did not improve with chemotherapy. After a second



Figure 6. Specimen. It shows total colon after surgical treatment. Due to adenocarcinoma at descending colon, subtotal colectomy was done.

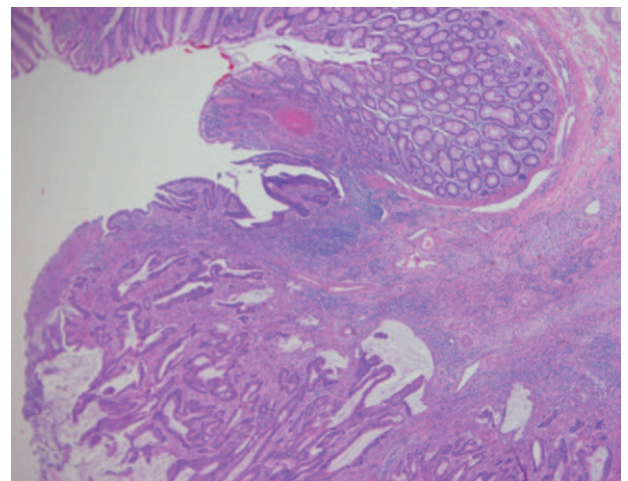


Figure 7. Histologic finding. It shows mucinous adenocarcinoma, poorly differentiated type (H&E $\times 400$). Tumor penetrated to the surface of the visceral peritoneum (pT4a), and regional lymph node metastasis was founded. H&E = hematoxylin and eosin.

round of chemotherapy, she and her parents denied further chemotherapy. Only pain control and conservative treatment were administered. She died 3 months later.

4. Discussion

Incidence of colorectal cancer is different in every country. Australia, New Zealand, Europe, and North America are estimated to have the highest incidence rates, whereas Africa and South Central Asia have the lowest incidence rate.^[1,6] The differences is due to the differences in dietary habits and environmental exposure.^[7] However, studies on major risk factors for colorectal cancer and global distribution in children are limited. In Korea, colorectal cancer is the third most common cancer in adults, but 5-year prevalence in patients aged ≤ 20 years is about 12 per 100,000 (2012 statistics).^[2] Furthermore, rectal cancer only occurs in patients aged above 15 years and the rate of colon cancer increases with age. Incidence rate of colorectal cancer in US pediatric population was about 1.3 per one million.^[8] In Japan, out of 700 pediatric malignant solid tumors annually, 1 to 3 were estimated to be colon carcinomas. In adults, despite the high incidence rate, mortality of colorectal cancer is low because of early detection and management.^[9] Therefore, early detection of colorectal cancer in young patients by extensive evaluation will contribute to more reported cases and will affect its incidence rate.

Symptoms of colorectal cancer in pediatric patients are not different from those seen in adults. Clinical manifestations differ depending on the tumor location; therefore, nonspecific gastrointestinal symptoms can be seen before the disease reaches an advanced stage.^[10,11] Underlying diseases and familial history of cancer could be vital information in suspecting colorectal cancer in adults. De novo carcinoma in a normal colon changes to malignancy faster than in a colon with chronic bowel diseases; therefore, fewer patients in the pediatric population have underlying diseases at first diagnosis, when compared with the adult population.^[12] In cases, chronic abdominal pain, hematochezia, or melena, persistent iron deficiency, and anemia can be related to colorectal cancer. Typical symptoms of colorectal cancer include vomiting, severe abdominal pain, and bloody stools; however, in children, the symptoms might be restricted to only altered bowel habits. Location of cancer can be a major factor in manifestation of specific symptoms. Similar to the first case presented, a right-sided cancer can manifest as mass, anemia, diarrhea, or intussusception. Left-sided cancer can cause bleeding, obstruction, or altered bowel habits, but similar to the second case, initial symptoms could be only a change in bowel habits with abdominal pain and diarrhea. Environmental factors can contribute to different prevalence in different age groups, but a major reason for the difference in the detection rate is the colorectal cancer screening system that has can potentially diagnose asymptomatic patients with colorectal cancers.^[13]

Unlike in adults, familial cancer history is not strongly associated with colorectal cancer in children. However, if patients who have familial cancer history have chronic symptoms, further imaging tests such as abdominal CT or endoscopy should be considered. Tumor makers can also be used to suspect the presence of colorectal cancer. Many serum markers are associated with colorectal cancer, especially CEA. However, serum markers, including CEA, have a low diagnostic ability when compared with radiologic examination due to low sensitivity (only 46%) and a possibility of false-positives,

including in other benign tumors. However, CEA levels of over 5 ng/mL predict a worse prognosis than lower levels.^[14]

As mentioned above, any symptoms indicative of colorectal cancer can be an indication for further imaging tests. Once colorectal cancer is suspected, barium enema, ultrasound abdominal examination, colonoscopy, abdominal CT, and therapeutic surgical resection can be performed. Barium enema is widely available and can be used in patients with symptoms of colorectal cancer.^[15] However, barium enema takes more time than abdominal CT and it is difficult to detect early states of cancer, which result in a low specificity. Colonoscopy is more useful because right ascending colon and transverse colon carcinomas are more common in pediatric patients. In Korea, pediatric endoscopic procedures can be commonly performed without general anesthesia at an endoscopy unit. In a comparative study in which 2527 patients were assigned to receive double contrast barium enema, the detection rate for colorectal cancer was significantly lower than that for CT or colonoscopy.^[16]

The Tumor Node Metastases staging system of the American Joint Committee on Cancer for International Cancer Control is the staging system of choice for colorectal cancer.^[17] To predict the prognosis, Duke and MAC staging systems are used together. In both these staging systems, stages A and B are curable. However, as in the second case presented, most pediatric patients are diagnosed with advanced-stage carcinoma with poor prognosis. Another reason for poor prognosis in pediatric patients is histological grading. In adults, mucinous adenocarcinoma accounts for less than 5% of cases. However, in children, more than 50% of patients are diagnosed with mucinous adenocarcinoma, which carries a poor prognosis.^[18] If a patient is diagnosed with colorectal cancer that is curable, complete resection is the ideal management. Left-sided tumors require subtotal colectomy, and right-sided tumors require extended hemicolectomy. According to TNM staging, adjuvant chemotherapy and radiotherapy are required, but these treatment regimens are controversial.

5. Conclusions

Mucinous adenocarcinoma, which has worse prognosis, accounts for higher incidence in young patients. Therefore, early detection can increase the possibility of curing cancer as described in the first case. In the second case, she showed the symptom of weight loss and anemia over 6 months. We believe that if earlier evaluation was performed, the patient might have lived. Since routine diagnostic colonoscopy in Korea is recommended for people aged >40 years, colorectal cancer in pediatric patients is detected late and in advanced stages. In Korea, pediatric endoscopy can be performed easily using deep sedation without general anesthesia at an endoscopic unit. In summary, if children present with persistent weight loss, bloody stools, or anemia, they need to undergo further imaging investigations or endoscopy.

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