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Basaloid (Cloacogenic) Carcinoma Mimicking Intraabdominal Abscess: Report of a Case and Review of the Literature

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Key Words
Cloacogenic carcinoma · High-molecular-weight cytokeratin · p63

Abstract
Cloacogenic carcinoma (CC, basaloid carcinoma) generally occurs around the dentate line, rarely it can arise from the other sides of the colon. There are only 5 cases of CC located outside the anal canal in the literature. The first occurrence of a CC presents as intraabdominal abscess. We describe a 23-year-old male patient who was admitted with fever and severe abdominal pain. Computed tomography imaging showed diffuse wall thickening about 10–11 cm above the rectosigmoid junction, intraabdominal abscess and a soft tissue lession covering the pelvis with a size of 8 × 8.5 cm including cystic necrotic areas. We performed Hartman procedure since the mass was nonresectable. Histopathological examination showed CC. In total, three times radiotherapy and a concurrent three-drug regimen of irinotecan, fluorouracil and folinic acid chemotherapy were administered for 6 weeks. As a result, the patient was lost because of multiple organ dysfunction syndrome that developed 3 months after radio-chemotherapy.

Introduction

Cloacogenic carcinomas (CCs) are relatively common neoplasms of the anal canal and are thought to arise from cloacogenic remnants at this site [1]. Histological and ultrastructural studies have indicated that these neoplasms may also arise from transitional cloacogenic epithelium lining the anal ducts or from the basal layer of anal

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squamous epithelium [1–3]. Outside the anal canal, it has been postulated that these tumors arise from cloacogenic embryologic remnants, squamous metaplastic epithelium, or totipotential basal cells [4–6].

We report a patient with CC of the recto-sigmoid junction presenting as intraabdominal abscess. For this reason, this case is the first reporting CC outside the anal canal in the literature.

Case Report

A 23-year-old man visited the emergency unit of our hospital with rectal bleeding, severe suprapubic abdominal pain, fever, poor general condition and fluid discharge from a median subumbilical incision. We learned that he had undergone an appendectomy at another center 8 months previously. Physical examination showed generalized peritonitis in the abdomen and his general condition was moderately dehydrated. The systolic and diastolic blood pressures were 80 and 50 mm Hg respectively, his pulse rate was 110/min, and his respiratory rate was 22/min. His body temperature was 39.3°C. Discharge of infected material from a median subumbilical incision was observed. Rectal examination revealed minimal bleeding. Laboratory results were as follows: blood urea nitrogen 65 mg/dl, creatinine 2.1 mg/dl, C-reactive protein 198 mg/l, potassium (K) 2.2 mEq/dl, and albumin 2.3 g/dl. Blood cell counts revealed leukocytosis (215,000/μl); his hemoglobin was 10.5 g/dl and the platelet count was 408,000/μl. Other serum parameters were within normal limits. We performed abdominal computed tomography (CT) with intravenous contrast, which showed diffuse wall thickening 10–11 cm above the rectosigmoid junction, and an 8 × 8.5 cm soft tissue lesion that included cystic areas in the pelvis (fig. 1, fig. 2). Multiple lymph nodes were seen in the abdomen; the biggest measured 3 × 2 cm. Air vesicles and images concordant with a fistula tract in the anterior abdominal wall were detected.

Laparatomy was performed. An 8 × 5 cm abscess was seen related to the fistula tract on the anterior wall of the abdomen. No intestinal content was observed in the abdomen. A 10 cm mass attached to nearby tissues that contained necrotic and caseated areas covered the distal sigmoid and proximal rectum. We analyzed frozen sections from the mass during the operation. Atypical cells were seen in the frozen sections. We performed abscess drainage because of these results. As resection was not possible, Hartman-type proximal end colostomy was performed and the operation was ended leaving a distal 'stubby'. As adenoid structures in patches were observed by histopathological examination, the differential diagnosis was made between poorly differentiated adenocarcinoma and CC. Immunohistochemically, staining with high-molecular-weight cytokeratin (HMWCK) (fig. 3), p63 antibody (fig. 4) and hematoxylin-eosin (fig. 5) confirmed the diagnosis of CC. On examination, CEA and CA 19-9 levels were normal (1.1 ng/ml and 2 IU/ml, respectively). Three weeks after discharge from the hospital, a 5-fluorouracil + folinic acid + irinotecan regimen was started. In total, three cycles of radiotherapy and a concurrent three-drug chemotherapy regimen (FOLFIRI; folinic acid, fluorouracil, irinotecan) were administered to the patient over 6 weeks. In CT scans 3 months after therapy, an increase in mass size and hydroureteronephrosis were observed. At the end of the fifth month, the patient's general condition deteriorated and sepsis developed. Subsequently, the patient died due to multiple organ dysfunction syndrome.

Discussion

In this case, failure in detecting the mass in the pelvis during laparatomy can depend on two reasons: either the mass was not formed at the time of the laparatomy or the mass could not be seen. Probably it was the colonic mass that caused symptoms of appendicitis in his first surgery. However, it is normal that colon carcinoma is not suspected initially in an 18-year-old patient presenting with abdominal pain without any family history, since without the presence of familial colon cancer or ulcerative colitis history, incidence of colon cancer is very low at <40 years of age. In this case, neither in family history nor in colonoscopic examination was any sign of polyposis coli present. Furthermore, because of aggressive progression but nearly normal CEA levels, no sign of liver metastasis, and no clinical answer to radiotherapy and chemotherapy, we thought that it was either

undifferentiated carcinoma arising from the colonic mucosa or anoderm originated CC which rarely occurs in the rectosigmoid area.

Review of the Literature

In the literature, most papers report positive reactions with HMWCK and p63 immunohistochemical antibodies in CC [7–10]. There are only 5 cases of CC located outside the anal canal in the literature. These are reported between 1977 and 2002. Strate et al. presented the first case of CC originating from the sigmoid colon and located outside the anal canal [4]. Hall-Craggs and Toker reported a second case originating from the sigmoid colon [5]. Ranaldi et al. reported a third case originating from the sigmoid colon [6]. Newell et al. reported a CC case originating from the splenic flexure of the colon with liver metastases [1]. Jaswal et al. presented a 24-year-old female patient with CC originating from the descending colon [11].

As a result, although CC generally occurs under the dentate line, rarely it can root from the other sides of the colon. It is difficult to diagnose it clinically. In suspected cases, the differential diagnosis can be done by immunohistochemical staining. Tumor size <2 cm, patient age <40 years, being poor in cells and having high fibroblastic activity are bad prognostic factors.

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Fig. 1. Spiral CT after administration of oral and intravenous contrast. Five-millimeter sections were obtained. The white arrow shows the fistula tract on the anterior wall of the abdomen.

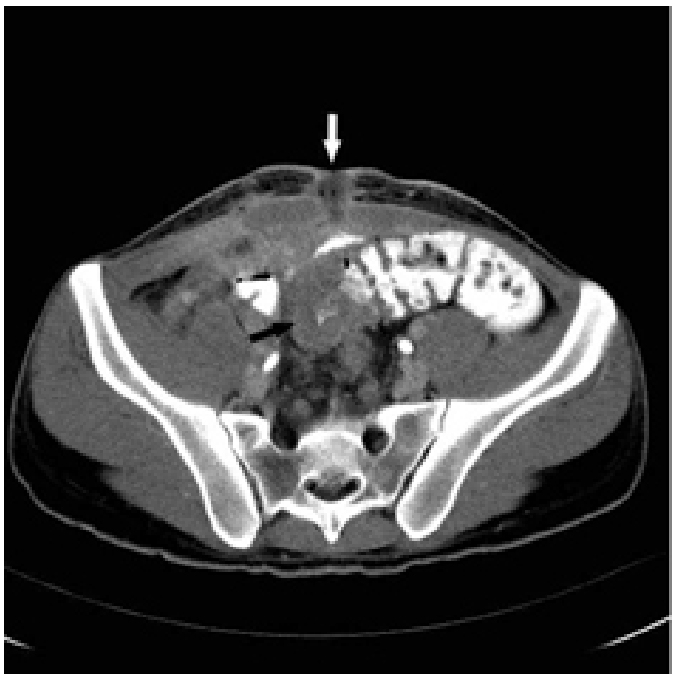

Fig. 2. The vertical arrow shows the fistula tract. The horizontal arrow shows the mass with necrotic areas which almost totally fills the pelvis.

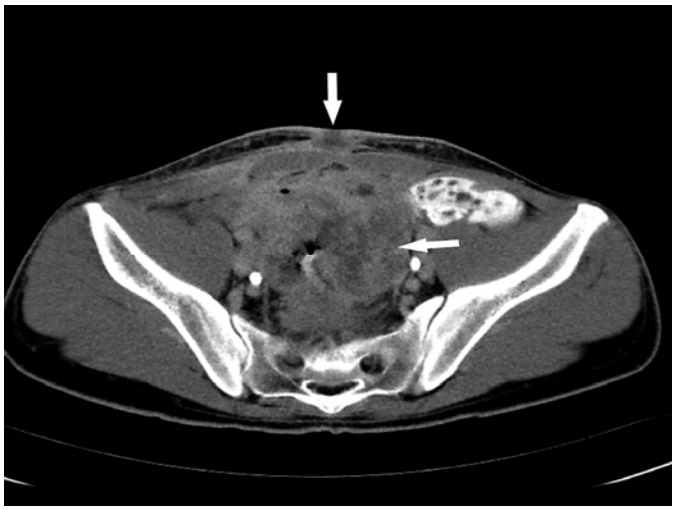

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Fig. 3. Positive staining with HMWCK in tumor cells (HMWCK antibody). Magnification ×200.

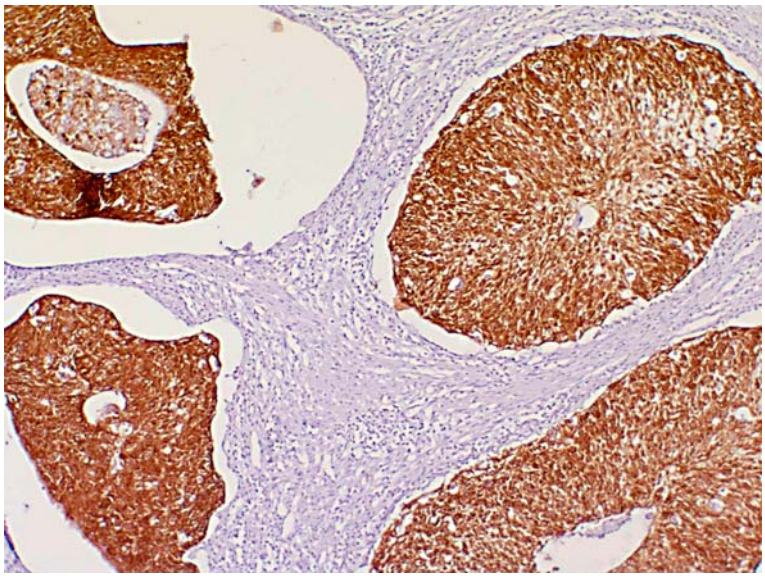

Fig. 4. Positive staining with p63 immunohistochemical antibody in tumor cells (p63 antibody). Magnification ×200.

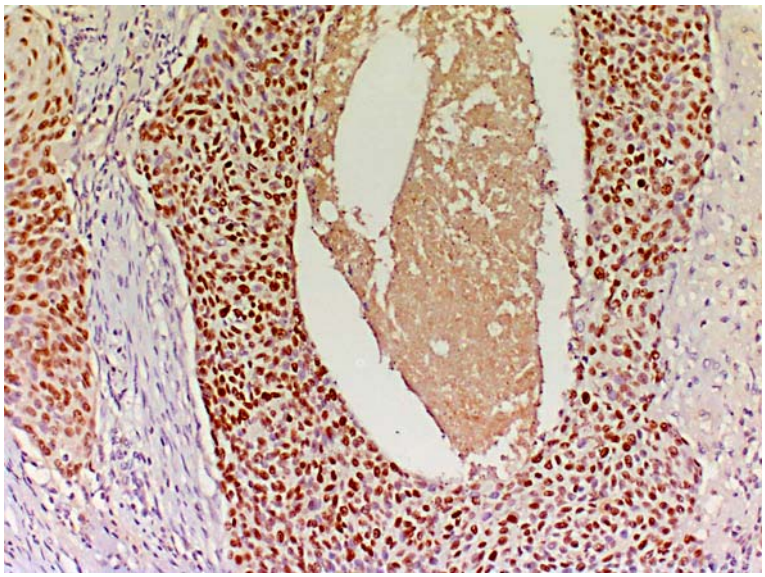

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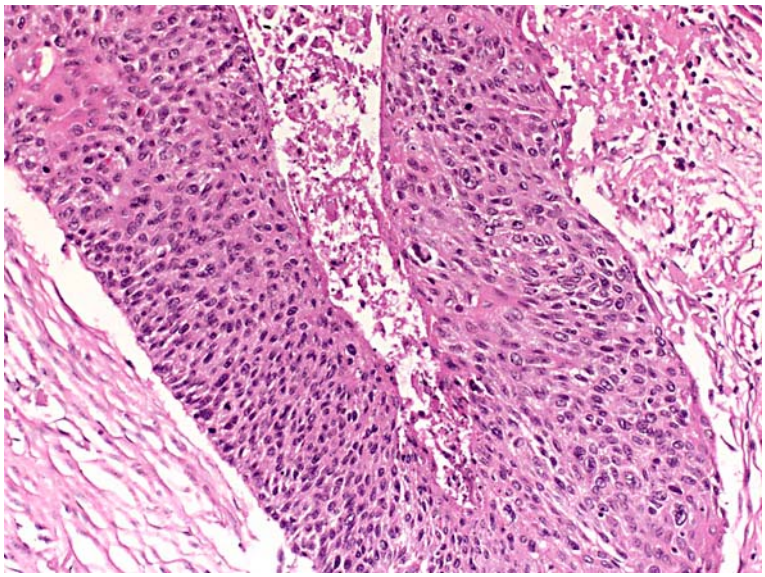
Fig. 5. Hematoxylin-eosin staining of the cloacogenic tumor. Magnification ×200.



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S. Akbulut wrote the draft of the manuscript and assisted in the surgical procedure. S. Akbulut performed the literature research. A. Sezgin examined the surgical specimen and wrote the pathological report. B. Cakabay performed the surgical procedure and designed study. C.A. Ozmen provided the radiological information.