Case Report

A Case of Spontaneous Colo-umbilical Fistula in Colon Carcinoma

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We herein report a case of a spontaneous fistula that developed from a carcinoma of the sigmoid colon that discharged through the umbilicus. A 72-year-old man was referred to our hospital for an examination of a foul odorous and purulent umbilical discharge. Six months prior to admission he complained of enuresis and ischuria for cystitis at the urological clinic of our hospital. The preoperative computed tomography and colonoscopy findings suggested a colo-umbilical fistula arising from the carcinoma of the sigmoid colon. At laparotomy, a 8-cm diameter mass, involving the anterior abdominal wall and the sigmoid colon, was found and it closely adhered to the upper aspect of the urinary bladder. An en bloc resection was thus performed. Its specimen showed an Borrmann 2-liked carcinoma of the sigmoid colon with an orifice of the colo-umbilical fistula, which was composed of a inflammatory tissue and no neoplastic cells were detected. This is the first case report of a spontaneous colo-umbilical fistula caused by a perforation of colon carcinoma, which demonstrated a pus discharge from the umbilicus. Carcinoma of the colon should thus be considered as a cause of an unexplained umbilical discharge.

key words : umbilical discharge, umbilical fistula, colon carcinoma

Introduction

Fistula formation has sometimes been reported as a complication in Crohn's disease¹⁾⁻³⁾ or embryonic anomalies⁴⁾. However, fistula formation complained as a colon carcinoma is uncommon. Indeed, fistula were found in the literature in less than 1% of carcinomas of the colon and rectum^{5),6)}. We recently encountered a unique case of a spontaneous colo-umbilical fistula secondary to carcinoma of the sigmoid colon. To our knowledge, no similar case due to colon carcinoma has been described previously. This case is herein presented, along with the proposed explanation of the mechanism in its formation.

Case report

A 72-year-old Japanese man presented with

a 3-day history of foul odorous and purulent umbilical discharge in June 1996. Six months prior to admission he complained of lower abdominal pain and enuresis and was treated with antibiotics for cystitis at the urological clinic of our hospital. During the six-month period prior to presentation, he had lost about 10 kg. On physical examination, his umbilicus showed a 2-cm diameter indurated mass with a central opening through which a moderate amount of purulent discharge was observed (**Fig. 1**). The laboratory data at admission revealed white-cell count of 17,500/cm³, hemoglobin of 11.0g/dl. Carcinoembryonic antigen was 12.2 ng/ml (normal under 5.0). Cystoscopy showed erosion of the posterior wall of the bladder, but not a fistulous opening. Computed tomography of the abdomen demonstrated a large low-attenuation mass involving the sig-

Fig. 1 Macroscopic appearance of the umbilicus. The umbilicus looks eroded, and feces is being excreted through its center.

moid colon extending from the umbilicus to the level of dome of the urinary bladder. Gas, as an air-fluid level, was present within the mass and these findings were considered to be most consistent with a penetrating carcinoma of the sigmoid colon. Colonoscopy revealed a easily bleeding mass, which completely occupied in the lumen of the colon at about 18 cm from the anus, but no fistula was identified. The biopsied specimen of the mass revealed adenocarcinoma (Group 5). These findings led to a preoperative diagnosis of colo-umbilical fistula due to sigmoid colon carcinoma and an operation was thus performed in June 1996. At laparotomy, a 8-cm diameter mass, involving the anterior abdominal wall and the mid loop of the sigmoid colon, was found and it closely adhered to the upper aspect of the urinary bladder. An en bloc resection, including a 20-cm section of the sigmoid colon, the affected abdominal wall

Fig. 2 The resected sigmoid colon. The arrow shows the orifice of the fistula to the umbilicus.

with the umbilicus and the invaded part of the urinary bladder, was undertaken. An end colectomy of the sigmoid colon was constructed, as the surgical margin was deemed positive due to the intimate involvement of the left ureter. A resected specimen of the sigmoid colon revealed an ulcerated tumor measuring 4 cm in greatest diameter, at the center of which the orifice of the colo-umbilical fistula was observed (Fig. 2). The fistulous canal was explored and found to extend to the middle umbilical ligament of the abdominal wall and then continued cranially to the umbilicus. A pathological examination showed the mass to be well differentiated adenocarcinoma of the sigmoid colon, which invaded directly to the serosal side of the urinary bladder (Fig. 3a). The fistulous canal was composed of collagenous, fibrous tissue and chronic perivascular infiltrates but no neoplastic cells could be detected (Fig. 3b). The tumor was graded as pathologic stage III (pT4N1M0) according to the TNM classification. The patient was discharged 30 days after the operation without any other problems and has been doing well so far.

Discussion

The umbilicus occasionally appears as an outlet of the fistula due to various causes because it is the thinnest part of the lower

Fig. 3 Photomicrograph : a A primary lesion of the sigmoid colon showing to be well differentiated adenocarcinoma. (H&E×100) b The fistula showing a fistulous tract with acute inflammatory infiltrates and granulation tissue. FA, fistulous tract. (H&E×40)

abdominal wall and thus offers the least resistance²⁾. The diversity of the umbilical discharge is partly attributable to the complex embryonic role of the umbilicus. Furthermore umbilical discharge can also be caused by various acquired diseases. Molderez et al.⁷⁾ classified umbilical discharge into either embryonic anomalies or an acquired pathology. The former group can be subdivided into 3 groups: 1) Non-regressed ductus omphalomesentericus, 2) Urachus remnants, and 3) Arteria umbilicalis remnants. The latter group can be subdivided to 4 groups: 1) Chronic inflammation of the umbilical basis, 2) Endometriosis, 3) Sister Mary Joseph's nodule, and 4) Other acquired anomalies. The present case was compatible with the acquired anomaly group. This group involves cases with dermoid cysts⁷⁾, sebaceous cysts⁷⁾, Crohn's disease^{1)~3)}, tuberculous peritonitis⁸⁾ and a perforated appendicitis⁹⁾. However, we failed to find a colo-umbilical fistula which developed as a sequela of perforated colon carcinoma in the available literature. On the other hand, regarding perforations of colorectal carcinoma, Welch et al.⁶⁾ categorized them into three types as follows: 1) an

acute perforation into the peritoneal cavity; 2) a subacute perforation, with a contained leakage of fecal material, resulting in abscess formation; 3) a subacute perforation into an adjacent structure, leading to a fistula or sinus tract. They reported that fistula formation to be uncommon since it occurred in approximately 1% of 2004 cases of colorectal carcinoma, and only 0.1% in which the fistula communicated with the skin. In Merrill's large series⁵⁾ from the Mayo Clinic only three patients had the colo-cutaneous fistula derived from colon carcinoma. External fistula formation caused by colon carcinoma is thus extremely rare, and indeed, almost all reports make no mention of its process. The mechanism of colo-umbilical fistula formation of the present case was not clear, but a few similar cases, such as Crohn's disease^{1,2)} and perforated appendicitis⁹⁾ have been reported. In these reviews, entero-umbilical fistula was considered to possibly cause a reopening the vestigial embryonic structure, due to the formation of an abscess near the urinary bladder. In the early fetal period, the umbilical cord includes the umbilical arteries and vein, the omphalomesenteric duct and the urachus. The omphalomesenteric duct connects to the intestine and the yolk sac, and the urachus carries fluid from the bladder to the allantois. Normally both ducts close off spontaneously at about 2 months gestation, thus forming solid cords⁴⁾. In this case, as the abscess increases the size, it might enter a vestigial embryonic structure and thereby force the pathway to the umbilicus to reopen because of its weak resistance. It is generally said that the dermis, being compact tissue, tends to resist neoplastic invasion⁵⁾. Retrospectively, this patient did complain of lower abdominal pain six months prior to presentation which was thought to be due to perivestigial abscess formation. That had developed due to a perfor-

ated colon carcinoma. Furthermore, a microscopic examination showed the fistula canal not to be constructed by neoplastic cells, but was instead composed of fibrous tissue and inflammatory cells. Merrill et al⁵⁾ expressed the opinion that the fixation of carcinoma of the colon adjacent structures was often due to inflammation rather than to a direct invasion of the malignant lesion and fistula creative type colon carcinoma also showed a more favorable prognosis than a freely perforated type when an extended resection en bloc with the constact areas was performed.

In conclusion, based on the above findings, perforative colon carcinoma should be included in the differentiated diagnosis of an unexplained umbilical discharge.

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