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■ BRIEF CASE REPORT ■

Adenocarcinoma Arising in a Colonic Duplication Cyst: A Case Report and Review of the Literature

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Duplications in the alimentary tract are uncommon congenital abnormalities that may occur anywhere from the oral cavity to the anus, with the ileum being the most common site.[1] Malignant change in a duplication is extremely rare.

Here, we report a case of adenocarcinoma arising in a colonic duplication cyst in a 23-year-old female and provide a review of the relevant literature.

CASE REPORT

A 23-year-old female patient was admitted with a palpable abdominal mass. The mass had been noted three years previously. On physical examination, a nontender and movable mass measuring approximately 7 cm was observed at the right lower quadrant.

The patient's serum carcinoembryonic antigen (CEA) level was 6.51 ng/mL (normal range, < 5.0 ng/mL), and that of cancer antigen 19-9 was 47.71 U/mL (normal range, < 37 U/mL).

Ultrasonography disclosed a hypoechoic cystic mass with an internal echogenic dot in the right lower quadrant. The echogenic dot was thought to be either an intraluminal secretion or necrotic debris (Fig. 1A). Computed tomography (CT) revealed a nonenhancing cystic mass measuring 7 cm, with linear calcification located adjacent to the medial side of the ascending colon (Fig. 1B). No enlarged regional lymph nodes, ascites, or other abnormalities were observed.

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A laparoscopic excision of the mass was performed. During the operation, the mass was observed to be attached to the ascending colon mesentery, having the blood supply by the right colic artery.

On gross examination, the cystic mass measured 8.6×6.4×2.9 cm, and its outer surface was smooth; mesenteric fat adhered to the outer surface. The maximum thickness of the cystic wall was 1.5 cm, and focal calcification was noted within the wall. The cyst contained brownish-colored mucoid material (Fig. 2A).

Microscopically, the cyst was found to have two well-organized layers of smooth muscle with an infiltrating adenocarcinoma forming irregular tubules (Fig. 2B, C). The tumor had invaded the pericystic mesenteric soft tissue and metastasized to eight of 16 mesenteric lymph nodes. Perineural and lymphovascular invasions were noted. Non-neoplastic glandular epithelium was not found; instead, a focal area of atypical columnar epithelium, which was compatible with low-grade dysplasia, was observed (Fig. 2D). On immunohistochemical study, tumor cells were positive for cytokeratin 7 (1:100, OV-TL 12/30, Dako, Glostrup, Denmark), cytokeratin 20 (1:100, KS20.8, Dako), CEA (prediluted, II-7, Dako), CDX2 (1:50, AMT 28, Novocastra, Newcastle upon Tyne, UK), and p53 (1:100, DO-7, Dako). The dysplastic epithelium was also positive for p53 (Fig. 2E).

Positron emission tomography-CT was performed after surgery and revealed no hypermetabolic areas other than the previous operation site. Two months after surgery, the patient started adjuvant chemotherapy.

DISCUSSION

Duplications are rare congenital anomalies that may occur

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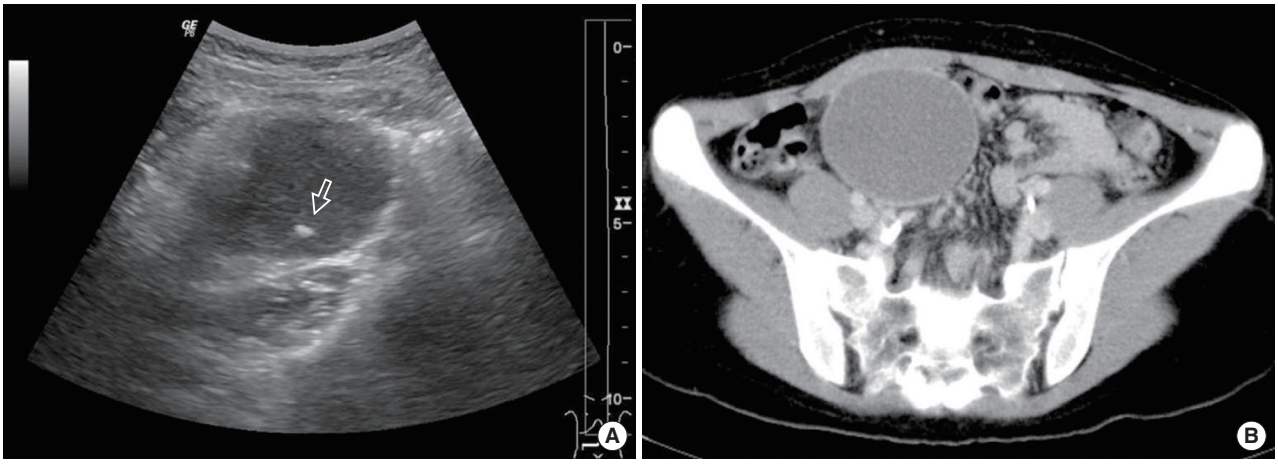

Fig. 1. Radiologic findings. (A) A hypoechoic mass with an echogenic dot (arrow) in the right lower quadrant is observed on ultrasonography. (B) A cystic mass with linear calcification in the mesentery adjacent to the ascending colon is observed on computed tomography.

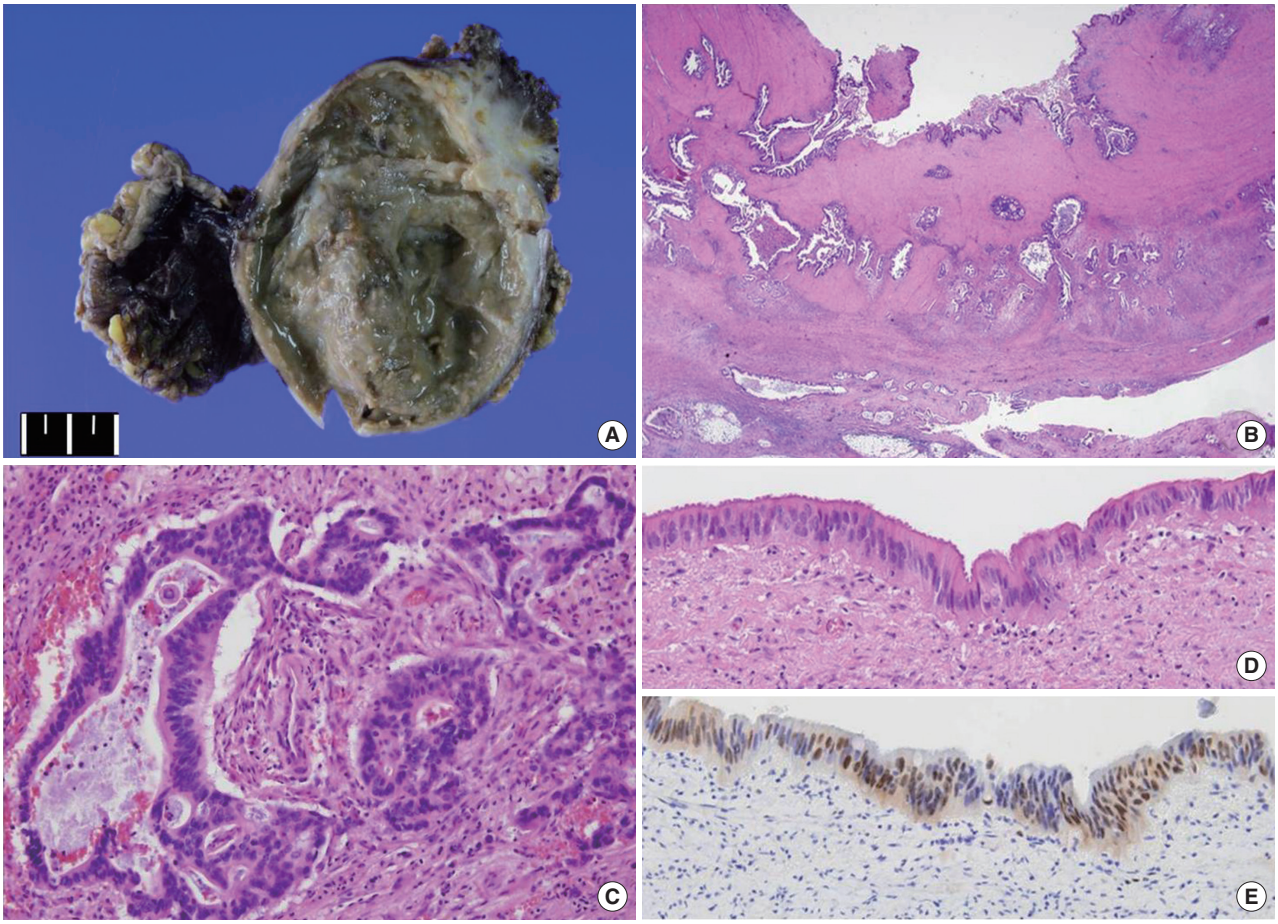

Fig. 2. Gross and microscopic findings. (A) The cystic mass measured 8.6×6.4×2.9 cm and contained brownish-colored mucoid material. (B) Full-thickness of two smooth muscle layers and adjacent mesenteric soft tissue are invaded by tumor cells. (C) The tumor is moderately differentiated adenocarcinoma. (D) A focal area of atypical columnar epithelium, compatible with dysplasia, is noted on the surface. (E) The dysplastic epithelial cells are positive for p53.

anywhere along the alimentary tract.[1] The most common site of involvement is the ileum. Colonic duplications comprise 4% to 18% of all duplications, and occur most often in the cecum. Strict morphologic criteria for the diagnosis of duplication have

Table 1. Clinicopathologic summary of cases with adenocarcinoma arising in a colonic duplication cyst

Case	Site	Age (yr)/ Sex	Symptom	Serum CEA	Size (cm)	Calcification	Depth of invasion	Treatment	Prognosis
1[2]	Cecum	61/F	Abdominal pain	ND	9	ND	Muscle layer	RHC, omentectomy, bilateral oophorectomy	NED
2[3]	Cecum	50/F	Lower abdominal pain	ND	8	ND	Pericystic mesenteric soft tissue	RHC	ND
3[4]	Cecum	40/F	Palpable mass	↑	11	N	Muscle layer	RHC	ND
4[5]	Cecum	41/M	Palpable mass	Normal	6	Y	Pericystic mesenteric soft tissue	RHC, CT	NED
5[1]	Ascending	41/F	RLQ pain	ND	4	Y	Thin fibrous wall (muscle layer)	Excision	NED
6[6]	Ascending	38/M	Palpable mass	↑	ND	Y	Capsule (muscle layer)	RHC	ND
7[6]	Ascending	59/F	Fever and lumbar pain	↑	ND	Y	Pericystic mesenteric soft tissue and right psoas muscle	Excision	ND
8[7]	Ascending	59/M	Melena due to adenocarcinoma in the ascending colon	ND	10	ND	Subserosa	RHC	ND
9[1]	Transverse	57/F	RLQ pain	ND	11	ND	Pericystic mesenteric soft tissue	RHC	NED
10[8]	Transverse	40/M	Palpable mass	ND	16	ND	Pericystic mesenteric soft tissue and omentum	Excision, CT	ND
11[4]	Descending	33/F	Palpable mass	ND	15	N	ND	Excision	ND
12[9]	Sigmoid	69/F	Epigastric pain	ND	15	Y	Pericystic mesenteric soft tissue and retroperitoneum	Excision	Died 41 mos later
13[10]	Sigmoid	72/F	Epigastric pain due to cholelithiasis	ND	3	ND	Subserosa	Sigmoid colectomy	ND
Present case	Ascending	23/F	Palpable mass	↑	8.6	Y	Pericystic mesenteric soft tissue	Excision, CT	NED

CEA, carcinoembryonic antigen; F, female; ND, no data available; RHC, right hemicolectomy; NED, no evidence of disease; N, no; M, male; Y, yes; CT, chemotherapy; RLQ, right lower quadrant.

been established: 1) attachment to the alimentary tract, 2) presence of smooth muscle layers, and 3) presence of lining epithelium resembling that of the alimentary tract. Our case met all of these criteria.

Malignant change in a duplication is very rare, and adenocarcinoma is the most common histologic type of malignancy found in these unusual cases.[1-10] However, squamous cell carcinoma, carcinoid tumor, gastrointestinal stromal tumor, and leiomyosarcoma have also been reported.

Thirteen cases of adenocarcinoma arising in a duplication of the colon have been reported to date in the English literature (Table 1).[1-10] Among the 14 cases, including the current case, four patients were male and 10 patients were female. The mean age at diagnosis was 48.8 years (range, 23 to 72 years) and our patient was the youngest. Some of the patients, including our patient, were relatively young, suggesting that the epithelium of the duplication has a high risk of carcinogenesis. The most common symptoms were abdominal pain and a palpable mass, and the mean size was 10.2 cm (range, 3 to 20 cm). Calcifications in the cystic wall or calculi in the lumen were common findings and were present in six of the eight cases where relevant data were available. Peripheral calcifications were present in three cases and calculi were found in four of six cases. Calcification in a duplication can be a worrisome finding because calcification is extremely rare in benign duplications.[6] The preoperative serum CEA level was available for five cases and was found to be elevated in four cases in which patients presented with advanced stage disease at the time of initial diagnosis. According to our survey, except for one case without relevant data, all cases had invasion beyond the muscular wall. Metastasis to a regional lymph node or to a distant organ was reported in three cases.[3,9] Although there was insufficient data on the duration of follow-up, five of six cases involved uneventful clinical courses. One patient died 41 months after diagnosis due to extensive metastasis.[9] No follow-up data was provided for the remaining eight patients. Due to its rarity and nonspecific symptoms, we thought that, in the present case, the diagnosis was made at an advanced stage. Calcification within the mass and elevated serum CEA level appear to be useful for preoperative prediction of malignant change in duplications.

In conclusion, intestinal duplication is an uncommon congenital abnormality, and malignant change in a duplication is extremely rare. Differential diagnosis of a cystic mass located in or adjacent to the gastrointestinal tract should include duplica-

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tion. When serum CEA level is high, and/or calcification within the mass is observed, the possibility of adenocarcinoma arising in a duplication cyst should be considered.

Conflicts of Interest

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

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