Abdom Imaging 23:135–137 (1998)

Abdominal Imaging

Adenocarcinoma arising in colonic duplication cysts with calcification: CT findings of two cases

Y. Inoue,[1] **H. Nakamura**[2]

[1]Department of Radiology, Minoo City Hospital, 5-7-1 Kayano, Minoo, Osaka 562, Japan
[2]Department of Radiology, Osaka University Medical School, Suita, Osaka, Japan

Received: 20 November 1996/Accepted: 24 December 1996

Abstract
We report the computed tomographic findings of mucinous adenocarcinoma with calcification arising from duplication cyst of the colon in two adult cases. In both cases, serum levels of carcinoembryonic antigen (CEA) were high. Differential diagnosis of intraperitoneal or retroperitoneal cystic tumors with mucinous density includes duplication cyst, and its malignant change should be considered when serum level of CEA is high.

Key words: Intestines, abnormalities—Intestinal neoplasmas—Calcification—Computed tomography.

Alimentary tract duplications are uncommon developmental anomalies that may occur anywhere in the gastrointestinal tract [1, 2], and they are commonly diagnosed in childhood. Their malignant change and calcification are quite exceptional [1]. We present the computed tomographic (CT) findings of adenocarcinoma arising in alimentary tract duplication with calcium deposits in two adult cases.

Case reports

Case 1

A 38-year-old man presented with a mass in the abdomen. Serum level of carcinoembryonic antigen (CEA) was 30 ng/ml (normal = <2.5 ng/ml). CT disclosed a mucinous cystic mass with calculi in the right lower abdominal space (Fig. 1A).

At surgery, a cystic tumor was found in the mesentery adjacent to the ascending colon. Tumor resection with right hemicolectomy was performed (Fig. 1B). The tumor was well encapsulated and was filled with a mucinous fluid. Microscopically, the inner surface of the capsule was lined by a papillary growth of mucin-producing malignant cells. There were several layers of smooth muscles in the wall (Fig. 1C). Microcalculi were seen in the cystic space. At some portions, malignant cells had infiltrated into the capsule. The pathological diagnosis was mucinous adenocarcinoma arising in the duplication cyst of the colon. The serum level of CEA became normal after surgery.

Correspondence to: Y. Inoue

Case 2

A 59-year-old woman developed a high fever and a right lumbar pain that did not respond to administration of antibiotics. She had undergone an appendectomy at 15 years of age but otherwise had been well.

The white cell count was 11,800/mm^3. CT disclosed a right retroperitoneal cystic mass extending to the right lumbar subcutaneous tissue. The diagnosis of retroperitoneal abscess was made at that time (Fig. 2). However, the laboratory data revealed a high serum level of CEA (17 ng/ml).

At surgery, a cystic tumor was seen in the right retroperitoneum, which adhered to the surrounding tissues. The tumor continued to the mid portion of the ascending colon through a narrow ductal structure. The tumor was resected with surrounding tissues. Microscopically, there were mucinous lakes containing malignant cells in the cystic spaces, and papillary growth of malignant cells lined the inner surface of the capsule containing smooth muscle layers, as in case 1. Malignant cells infiltrated into the surrounding tissues including the right psoas muscle. The pathological diagnosis was infected mucinous adenocarcinoma arising in the duplication cyst of the colon. The level of serum CEA returned to normal after surgery.

Discussion

Alimentary tract duplications are usually contiguous with the bowel and assume spherical or tubular structures. They may or may not communicate with the lumen of the bowel. Histologically, the inner surface is lined with a mucosa similar to some portion of the alimentary tract, and the wall is composed of several layers of smooth muscle [1–4].

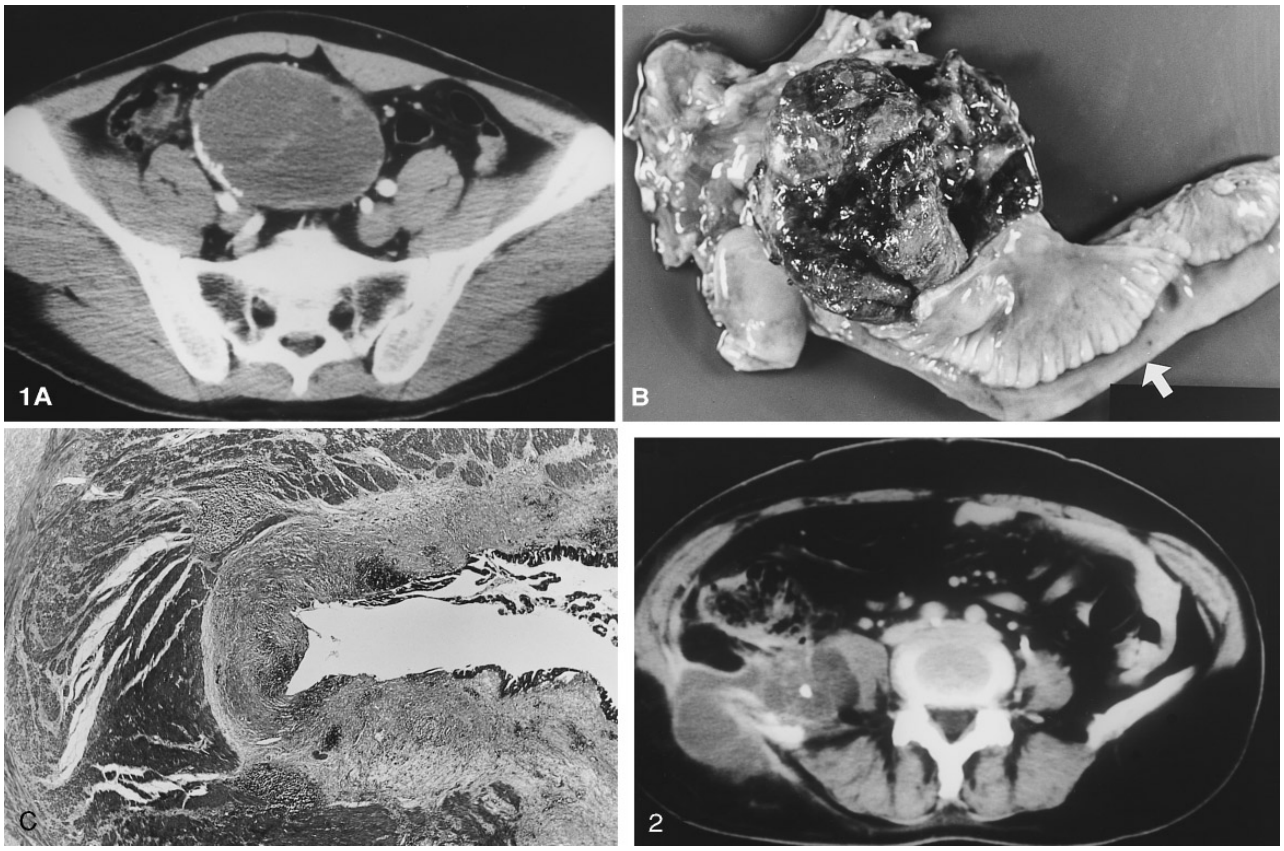

Fig. 1. Case 1. **A** Enhanced CT shows a cystic mass with an irregularly thickened capsule and septum. Many tiny calculi are seen along the capsule. The attenuation value within the cyst was 34–36 HU. **B** Resected specimen shows a cystic mass that is open through the anterior wall. The tumor is contiguous to the ascending colon (*arrow*). **C** Photomicrograph demonstrates the smooth muscle layers in the tumor capsule. The inner surface is lined by mucin-secreting malignant cells. Hematoxylin and eosin, ×3.3.

Fig. 2. Case 2. Enhanced CT shows a retroperitoneal cystic tumor extending to the right posterior wall of the abdomen. A central calculus in the tumor appears as an enterolith. The attenuation value in the mass is 25–32 HU.

In our survey, we found 16 cases in the English literature of malignant tumors arising from alimentary tract duplications [5–18]. The 18 cases, including the present two, consisted of six men and 12 women, ranging in age from 33 to 65 years. Although the colon is the least common site of duplication (6–8%) [1, 3, 4], tumors in 12 of the 18 cases (67%) occurred in colon or rectum. Adenocarcinoma occurred in 14 cases, and squamous cell carcinoma in four.

Rice et al. [15] described CT findings of two cases with carcinoma in duplication cyst, in which both cases demonstrated a thin-walled and noncalcified cyst with a soft tissue density nodule representing a focus of adenocarcinoma. In our cases, such a malignant focus was not defined, and malignant cells totally replaced the inner surface of the cysts infiltrating into the wall. In case 2, the cyst wall was poorly defined due to its perforation into the surrounding tissue and infection. The infection may be accounted for by the connection of the cystic space with the colonic lumen.

In the present two cases, the attenuation values in the cysts were mucinous. Mucinous density in cystic tumors may imply the existence of mucin-secreting epithelium [19], and therefore the possibility of adeocarcinoma should be considered, especially when serum level of CEA is high.

Calcification in alimentary duplication is extremely rare. We found eight cases of calcification in alimentary tract duplication in the English literature [9, 20–24]. In five cases, calcification was seen in the wall [9, 20–22]; in three cases, enteroliths were seen in the lumen [22–24]. In case 1, calculi were seen in the mucinous lakes. In case 2, central calculus looked like an appendicolith, which was impossible because of the history of appendectomy, and may be enterolith in the duplication cyst. In our survey, we found only one previous case with concomitant occurrence of malignancy and calcification in alimentary tract duplication, in which calcification was seen beneath the epithelium and in necrotic tumor [9].

Differential diagnosis of the intraperitoneal or retroperitoneal cystic tumor should include alimentary tract duplication whether or not it contains calculi. Its malignant change should be considered when serum level of CEA is high.

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