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Alpha-Fetoprotein Producing Cancer of The Colon: A Case Report and Literature Review

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ABSTRACT

We recently experienced a 57-year old woman with pure yolk sac tumor in the colon. Colonoscopy biopsy showed poorly differentiated adenocarcinoma. A right hemicolectomy was performed. The tumor showed a polymorphous pattern, in which the tumor cells grew in solid sheets with papillary and glandular structures; In addition, reticular-microcystic pattern and Schiller-Duval body were also observed. Alpha-fetoprotein using different immunochemical stains were positive, including Glypican-3, HepPar1, AFP, CDX-2; beta HCG was negative, ruling out the possibility of containing choriocarcinoma. These morphologic and immunohistochemical characteristics confirmed the diagnosis of yolk sack tumor. Here, we describe a case of pure yolk sac tumor, which may contribute to a better understanding of this tumor.

Keywords: Alpha-fetoprotein, Colon, Immunochemical stains, Yolk sac tumor.

INTRODUCTION

No tumor marker is specific for a particular disease. Initially, elevated serum alpha-fetoprotein (AFP) signals the presence of hepatocellular carcinoma (HCC) or yolk sac tumor (YST) in patients. Later, elevation of serum AFP was also found in cancer from the stomach[1,2], esophagus [3], lung[4] and omentum [5]. There had been a plethora report on alpha-fetoprotein producing gastric carcinoma (AFP-GC) in the literature, predominantly from Japan and China [6,7], a few from Taiwan and other part of the world. Second to AFP-GC was alpha-producing colorectal cancer (AFP-CRC). Nevertheless, the reported AFP-CRC was traditional adenocarcinoma combined with choriocarcinoma or yolk sac tumor. Recently, we encountered a patient who was thought to have AFP-CRC, histological examination after surgery turned out to be pure yolk sac tumor, which is extremely rare. Herein, we report this case and review the literature.

CASE REPORT

A 57-year-old woman visited our outpatient clinic complaining of poor appetite、abdominal discomfort、nausea and bloody stool for the past two weeks. . Laboratory examination revealed anemia, with Hemoglobin level of 11.1 g/dl, HCT of 32.7%. The white blood cells and the platelet count were within normal limits. The blood chemistry profile showed nothing of note. However, her tumor markers were abnormal; Alpha-fetoprotein (AFP) was 1319 ng/ml (normal <7 ng/ml). Serum levels of carcinoembryonic antigen (CEA) and human gonadotropin (HCG) were within normal limits. She underwent a colonoscopy examination, a large tumor was found at the ascending colon near the hepatic flexure. Biopsy was done, and the diagnosis of poorly differentiated adenocarcinoma was rendered. Subsequently, right hemicolectomy and lymph node dissection was then performed. The tumor was adhered to the duodenum and pancreas. Dissection was performed with somewhat difficult. There was no metastasis in the liver. Her Serum level of AFP was 370 ng/ml 9 days after surgery. Port A tube was installed, unfortunately, she went to a medical center for further therapy.

Histopathology

The postoperative specimen consisted of a segment of colon attached with an appendix. On opening, there was a fungating tumor with central ulceration, measuring 4x3x2 cm, four cm from the proximal resection margin **(Figure 1)**. The tumor was grayish white in color, and apparently has invaded through the muscle layer to the serosa. Viewing from the back side, the serosa appeared hemorrhagic with fibrous adhesive bands, indicating adhesion to the adjacent organs. Sections from the tumor displayed a polymorphous appearance, i.e. the tumor was composed of a solid sheets with glandular or papillary structure **(Figure 2)**. The solid sheets consist of closely packed cells with eosinophilic or clear cytoplasm. In addition, they exhibited a variety of patterns characteristic for the YST: glandular, papillary, clear cell, hepatoid and reticular microcystic. A few Schiller-Duval bodies were observed **(Figure 3)** .Immunohistochemical stainings (IHCs) **(Figure 4)** showed the following results: HepPar 1 3+, diffuse; Glypican-3+, focal; AFP 3+, focal; CDX-3 +, diffuse. HCG was negative (not shown). Histological evaluation, in conjunction with IHC studies, we made this case as yolk sac tumor. Because the tumor invaded through the visceral peritoneum, with six lymph nodes positive out of seventeen examined. There was no radiological evidence of liver metastasis. This tumor was staged as pT4aN2a. Later, we submitted the whole tumor for histological examination, no conventional adenocarcinoma or choriocarcinoma were found which led us to diagnose this tumor as pure yolk sac tumor of the colon.

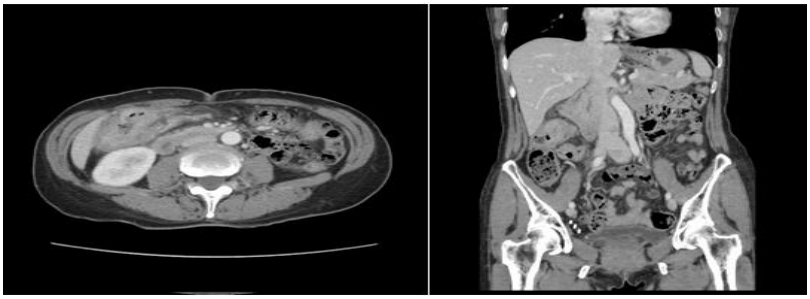

Figure 1: Computer tomography of coronal view (left) as well as axial view (right) displaying thickening of the bowel wall indicating a large tumor partially obstructing the lumen.

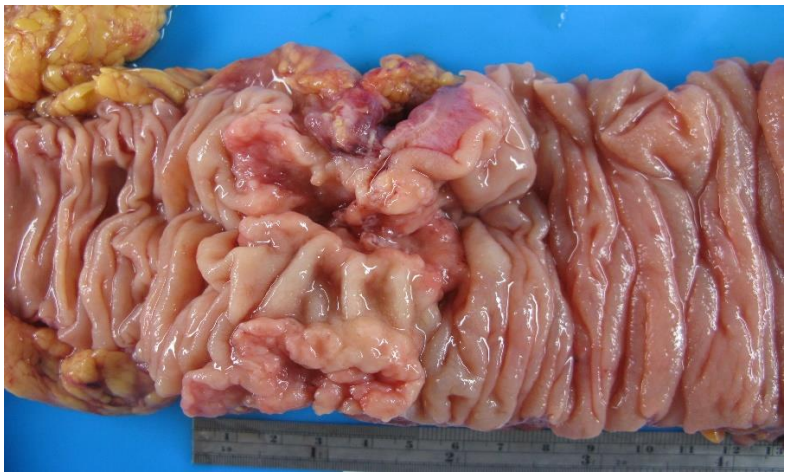

Figure 2: The resected specimen displayed a fungating tumor with central ulceration at the ascending colon near the hepatic flexure.

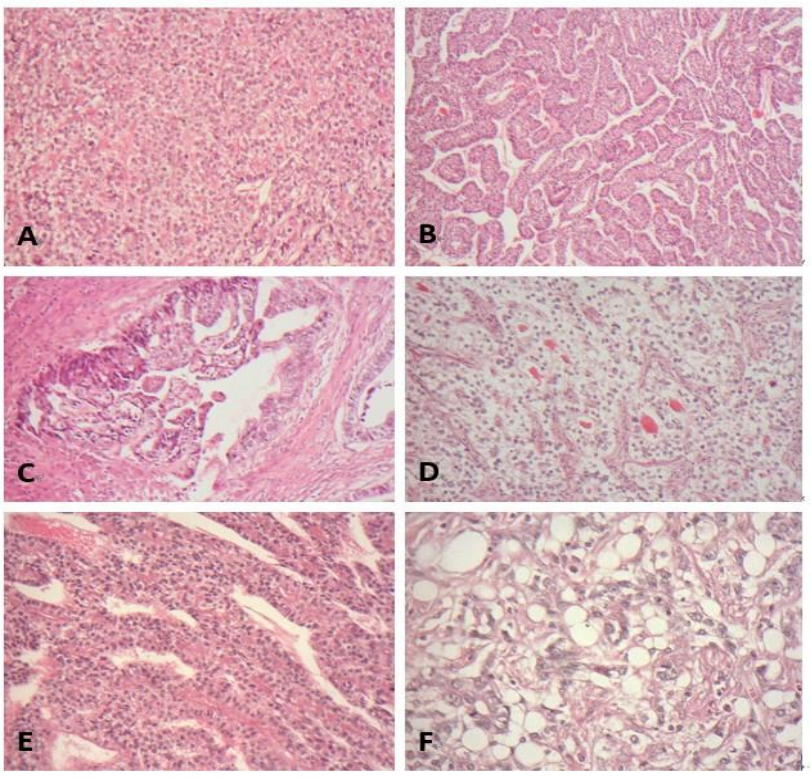

Figure 3: Histological sections of the tumor displayed polymorphous pattern (H & E stains, x200). (A) Solid pattern. (B) Glandular pattern. (C) Papillary configuration. (D) Clear cell pattern. (E) Hepatoid pattern. (F) Reticular-microcyst.

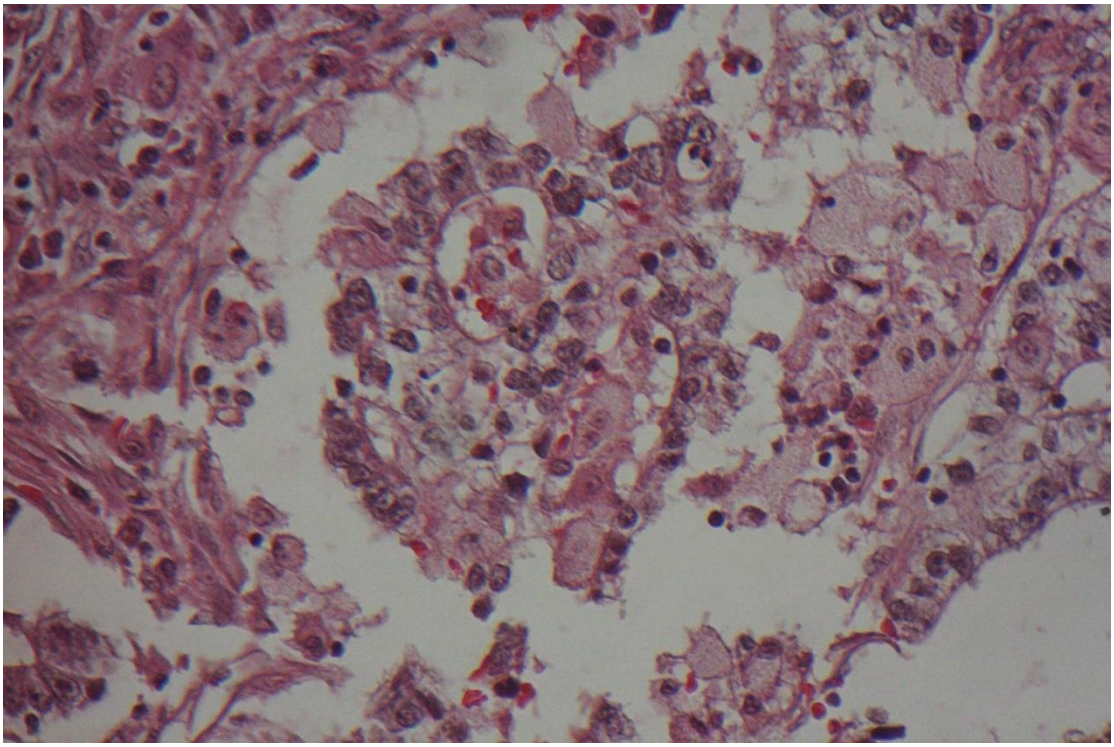

Figure 4: Schiller-Duval body. (H&E stain, x400).

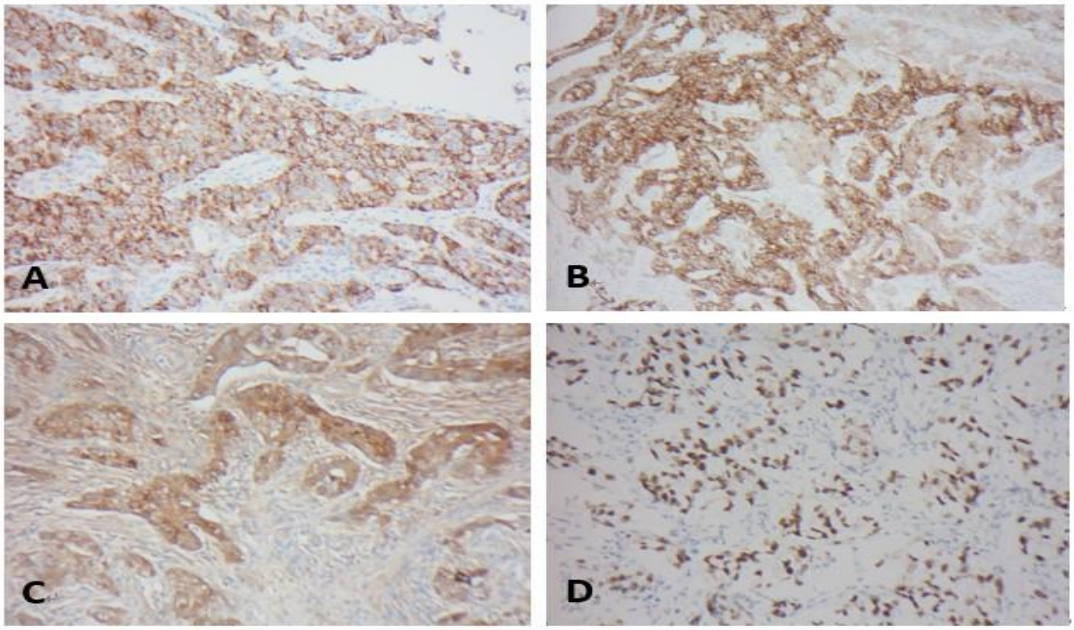

Figure 5: Immunochemical stains, x200. (A) HepPar 1 3+, diffuse. (B) Glyican-3: 3+, focal. (C) AFP:3+, focal. (D) CDX-2:2+, diffuse.

DISCUSSION AND LITERATURE REVIEW

AFP-GC accounts for the majority of AFP-producing cancers. AFP-GC is considered as the subtype of gastric cancer, because it has propensity to metastasize to the liver and lymph node, resulting in poor prognosis. AFP-CRC behaves aggressively, similar to that of AFP-GC. The cellular and molecular mechanism responsible for their poor prognosis are not clearly understood. An early report indicated that AFP had a suppressive effect on lymphocyte transformation [8]. Another report indicated that AFP could enhance proliferative activity and increase angiogenesis due to positive expression of vascular endothelial growth factor VEGF [9]. In a study in 2000, Amemiya et al, [10] found that hepatocyte growth factor (HGF) and c-met expression was higher in AFP-GC than that in AFP negative gastric cancer. Through the HGF and c-met pathway, tumor growth was promoted.

AFP-CRC is extremely rare, since Nakajima et al reported the first case of AFP-CRC in 1985 [11], only a few cases have been sporadically published in the literature [12], the largest series was from Ren et al [13]. They analyzed 20 patients on clinicopathological features and prognosis. None of their cases had yolk sac tumor. According to Otani et al [14], only four cases of colorectal adenocarcinoma with YST component have been reported in the English literature up to 2022, but they were traditional adenocarcinoma combined with YST, or with YST and choriocarcinoma . To the best of our knowledge, our present case is the first report of pure colonic YST. It is worth mentioning that endodermal sinus and yolk sac tumor are used interchangeably. The pathogenesis of the yolk sac tumor in the colon cancer still remains unknown. One theory proposed a retrodifferention mechanism of mature cells into primitive pluripotential cells [15]. A case report by Otani et al [14] supported this theory. In their patient, the cells from the original adenocarcinoma and yolk sac tumor in the recurrence shared mutation in APC and TP53 genes. The second theory was that of the migration misplacement. During embryogenesis, the primitive gonadal cells migrated from the cranial cavity to the external genitalia. During their migration, some primitive germ cell might misplace at the migration pathway. These cells could transform into germ cell tumor [15]. Currently, there is no standard guideline to treat AFP-CRC patients with germ cell component. In the literature, they were usually treated according to traditional adenocarcinoma, receiving adjuvant chemotherapy after resection. It was promising that some patients could benefit from preoperative chemotherapy [16-20].

In conclusion, we reported the first case of pure yolk sac tumor in colon, in which primary tumor resection and lymph dissection were performed. It was unfortunate that our patient was transferred to a medical center for further therapy. We missed the opportunity to gain the knowledge about treating pure yolk sac tumor of the colon.

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