

Unusual abscess masquerading as poorly differentiated adenocarcinoma of the colon showing characteristics of choriocarcinoma

Antigony Mitselou^a, Anna Varouktsi^b, Stamatis S. Papadatos^c , Eugenia Balasi^a, Evangeliki Lampri^d, Konstantinos Katsanos^e, Vasiliki Galani^f

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ABSTRACT

Extragenital non-gestational choriocarcinoma (ENC) is an uncommon malignant tumor occasionally found in the gastrointestinal tract. ENC is characterized by a biphasic tumor growth with distinct areas of adenocarcinoma and choriocarcinoma differentiation. Primary choriocarcinoma of the colon is extremely rare, with only 21 cases reported in the literature. Most of the perforation of colorectal cancers occurs in the abdominal cavity, while abdominal wall abscess is rare; the psoas abscess associated with colon carcinoma is even less observed. Herein, we report the case of a 61-year-old female with poorly differentiated adenocarcinoma of the ascending colon and sigmoid, with choriocarcinomatous differentiation, masquerading a psoas abscess formation. Unfortunately, despite the aggressive therapy, the patient's disease rapidly progressed, and she died within 2 months after the diagnosis. The typical morphological pattern, immunohistochemistry, and its correlation with serum β -human chorionic gonadotropin enabled a correct diagnosis.

Keywords

Adenocarcinoma; Choriocarcinoma; Abscess; Colon; Extragenital Nongestational Choriocarcinoma.

INTRODUCTION

Choriocarcinoma is a rapidly invasive, aggressive, and widely metastatic human chorionic gonadotropin (hCG)-producing neoplasm, which develops from a preceding hydatidiform mole. Occasionally, choriocarcinoma is generated from the ovary and the testis, as a component of germ-cell tumors.¹ Extragenital non-gestational choriocarcinoma (ENC) is uncommon and mostly arises in the midline structures, such as the mediastinum, thymus, and the retroperitoneum.¹⁻³ However, ENC may also arise in non-midline organs,

such as the stomach, lung, liver, pancreas, the small and large bowel, cervix, ureter, and urinary bladder.¹⁻⁴ In the gastrointestinal tract, the stomach is the most frequently involved organ. Histologically, in the digestive tube, the neoplasm is characterized by a biphasic tumor growth with separated areas of adenocarcinomatous and choriocarcinomatous differentiation.⁴ Primary choriocarcinoma of the colon was first described by Park and Reid³ in 1980, and since then only 20 cases have been reported.⁵

^a University of Ioannina School of Health Sciences, Department of Forensic Medicine. Ioannina, Greece.

^b Ippokratia Hospital, Department of Internal Medicine. Thessaloniki, Greece.

^c National and Kapodistrian University of Athens, Medical School, 3rd Department of Internal Medicine. Athens, Greece.

^d University of Ioannina, Cancer Biobank Center. Ioannina, Greece.

^e University of Ioannina School of Health Sciences, Department of Internal Medicine, Division of Gastroenterology. Ioannina, Greece.

^f University of Ioannina, School of Health Sciences, Department of Anatomy, Histology and Embryology. Ioannina, Greece.



Abscess formation associated with colon cancer is a rare complication; even more uncommon is the abscess' formation within the psoas associated with colon carcinoma.^{6,7} Herein, we present the case of a female patient with rapidly progressive poorly differentiated adenocarcinoma of the colon with choriocarcinomatous differentiation uncovered as an abscess in the right lower abdomen. We discuss the clinical and pathological features with a review of the literature.

CASE REPORT

A 61-year-old woman attended our hospital for investigation of pyrexia, abdominal pain, anorexia, changes in bowel habits, weakness, and weight loss. The computed tomography (CT) scan demonstrated a low-density area in the swollen iliopsoas muscle and abdominal wall, a thickened cecum wall, and a swollen appendix. The differential diagnosis included plastron appendicitis and perforated colon malignancy. Ten days after admission, a follow-up CT scan revealed the enlargement of the right iliopsoas muscle. Fluid was collected in the pouch of Douglas. A colonoscopy revealed a large mass in the cecum, and a second obstructive mass in the sigmoid colon. The preoperative serum β -hCG measurement was not performed due to the lack of suspicion of choriocarcinoma. At laparotomy, multiple nodules were found disseminated in the peritoneum, along with abscess formation in the retroperitoneum and iliopsoas muscle, which were incised and opened. Right hemicolectomy and sigmoidectomy were performed. Liver metastases were also found. The pancreas and pelvic organs were normal. The postoperative serum β -hCG measurement was 112,000 mIU/mL (normal value <2 mIU/mL).

The pathologic findings showed that cecal and sigmoid tumors invaded the intestinal serosa and was composed of poorly differentiated adenocarcinoma with a second component showing syncytiotrophoblastic and cytotrophoblastic-like cells, along with necrosis and hemorrhage (Figure 1 and 2). Immunohistochemical staining was positive for hCG (Figures 3), CA19-9, and cytokeratin 9. The tumor was partially positive for cytokeratin 20, EMA and CEA, whereas it was negative for cytokeratin 7. Metastasis was found in the liver, peritoneum, and 21 excised lymph nodes. The tumors were classified as T4, N2, M1 (TNM classification) and staged as Duke's D (Stage IV). The patient was treated with systemic polychemotherapy. However, 2 months after the initial diagnosis, the patient died.

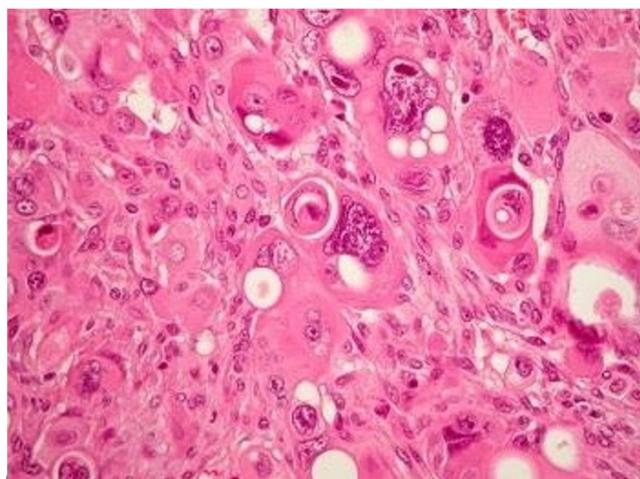


Figure 1. Photomicrographs of the tumor. Choriocarcinoma composed of highly pleomorphic cells with cytotrophoblast cytoplasm and multinucleated giant cells (H&E, 400X).

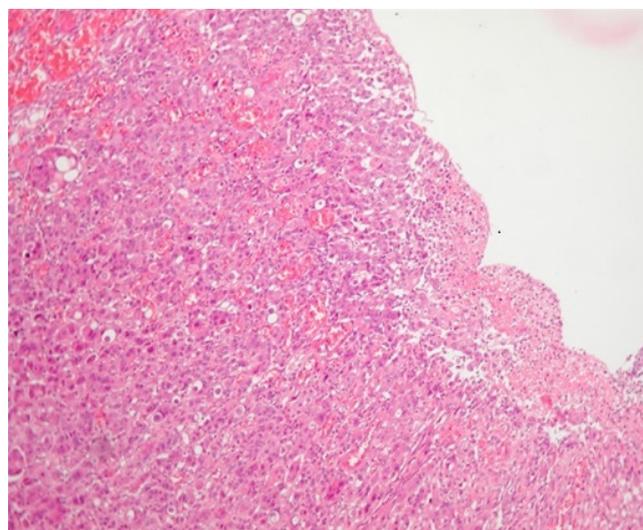


Figure 2. Photomicrograph of the tumor showing choriocarcinomatous and adenocarcinomatous component closely adjoining each other (H&E 100X).

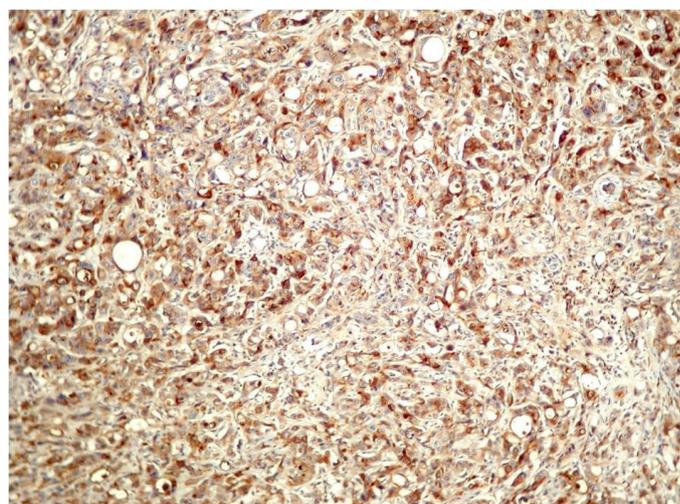


Figure 3. Strong immunoreactivity of β -human chorionic gonadotropin (β -HCG) in cancer cells (400X).

DISCUSSION

Abscess formation has been reported to occur in 0.3% to 4% of colonic carcinomas; however, it is the second most common presentation of perforated lesions.⁶ Colon carcinoma-related psoas abscess is very rare with scant cases reported in the literature.⁷ The preoperative diagnosis of abscess related to colon cancer is difficult yet crucial. Fever, pain, palpable mass, and leukocytosis are some clues that may guide the diagnosis of abscess. The association of abscess and

malignancy should be suspected in patients with an atypical history of prolonged symptoms, a palpable mass, weight loss, or anemia, as observed in our case.^{6,7}

Choriocarcinoma usually arises in trophoblastic tissue. Extragonadal, non-gestational choriocarcinoma is rare, but is occasionally found in the gastrointestinal tract, most frequently in the stomach.^{2,3} Primary colorectal choriocarcinoma was reported in 1980 for the first time, and to date, 22 cases (including the present one), have been reported in the literature (Table 1).^{3,5,8}

Table 1. Reported cases of colorectal choriocarcinoma (Modified from Oh et al.⁵)

Author	Sex/Age (yr)	Primary colon tumor location	Primary colon histology	Metastases	Metastatic tumor histology	Serum β -hCG (mIU/mL)	Treatment	Survival time (mo)
Park and Reid ³	F/49	Sigmoid	A+C	Liver, lung	C	NA	Palliative resection	4
Nguyen ⁴	M/74	Sigmoid	A+C	Liver	C	400 (bo)	Laparotomy	3
Ordóñez and Luna ⁹	F/35	Ascending	A+C	Liver, lung	C	1,612 (ao)	Right hemicolectomy	2
Kubosawa et al. ¹⁰	F/50	Sigmoid	A+C	Liver, lung	C	230,000 (bo)	Hartmann's procedure	4
Metz et al. ¹¹	F/42	Sigmoid	A+C	Liver, lung, spleen	C	154,000	Laparotomy	1
Lind and Haghghi ¹²	M/42	Ascending	C	Liver, lung, spleen	C	610,000	Palliative resection	1
Ostör et al. ¹³	F/28	Rectum	A+C	Liver	C	16,500 (bo)	Arterial resection, EMA/CO	1
Tokisue et al. ¹⁴	F/29	Rectum	A+C	Lung, brain, vagina	NA	49,000 (b CTX)	EMA, arterial resection	10
Oh et al. ¹⁵	M/69	Sigmoid	A+C	Liver, lung, thyroid, brain	C	78	Colectomy, MTX	15
Kim et al. ¹⁶	M/59	Cecum	A+C	NA	NA	NA	NA	NA
Kiran et al. ¹⁷	M/68	Rectum	A+C	Liver	NA	700,000	Hartmann's procedure	NA
Le et al. ¹	M/73	Ascending Descending	C	Lung, brain, kidney	NA	146,000	No	15 days
Verbeek et al. ¹⁸	F/54	Rectum	A+C	Liver, lung	NA	6,831 (ao)	Palliative resection, CTx	8
Jeong et al. ¹⁹	M/52	Rectum	A+C	Liver, lung	NA	4,224	Low arterial resection	47 days
Kourda et al. ⁸	F/42	Sigmoid	A+C	Liver	C	8,235 (ao)	Palliative resection	1
Froylich et al. ²⁰	F/57	Descending	C	Lung, bone, brain	C	13,000	Colectomy, VP16	16
Harada et al. ²¹	F/58	Sigmoid	A+C	No	No	2,420 (bo)	Hartmann's procedure	60
Jiang et al. ²	M/38	Ascending	C	Liver	NA	10,000 (ao)	Colectomy, CTx	6
Maehira et al. ²²	M/68	Sigmoid	A+C	Liver	NA	1.4 (ao)	Sigmoidectomy, FOLFOX, FOLFIRI	9
Mardi et al. ²³	F/54	Rectum	A+C	No	No	5568	Radical resection, 5-FU	50 days
Our case	F/61	Cecum Sigmoid	A+C	Liver, peritoneum	NA	112000	Colectomy, sigmoidectomy, CTx	2

A = adenocarcinoma; ao = after operation; b-CTX= Beta- CTX; bo = before operation; C = choriocarcinoma; CTx, chemotherapy; EMA/CO, chemotherapy; F = female; FOLFIRI, chemotherapy; FOLFOX, chemotherapy 5- FU = Fluorouracil; hCG = beta-human-chorionic gonadotropin; M = male; mo= months; MTX =Metaxin; NA = not available; No= without; VP16 =Herpes simplex virus protein vmw65; yr =years.

The patients' ages ranged from 28 to 74 years (mean age 52.86 years). There were 13 (59.10%) females and 9 (40.90%) males. The sigmoid colon (9 cases [40.90%]) was most frequently involved. Regarding the histological findings, adenocarcinoma was accompanied in 18 cases (81.8%), and at the time of surgery, most patients had metastasis (19 cases [86.3%]). The lymph nodes were the most common metastatic site, followed by the liver, lung, and peritoneum. The median survival period was 7 months (range 0.5-60 months). Almost all the reported patients (16 cases [72.7%]) died within 1 year of diagnosis. Only one case was reported to have achieved long-term (60 months) relapse-free survival.²¹

The pathogenesis of trophoblastic differentiation in non-gestational and non-germ-cell tumors is not well understood.³ There are five important theories that link choriocarcinoma with "embryonic remnants" and suggest that the failure to complete the migration of germ cells to the gonadal tissue and undergoing their apoptosis, may contribute to malignant transformation and, finally, transformation into the choriocarcinoma.

In addition, many studies^{1,3,10,14,18,21} demonstrated that a tumor consisting of mature tissue elements may undergo retro differentiation or dedifferentiation, resulting in cells with different morphological features. The dedifferentiation theory is considered the most reasonable.

Recently, molecular studies using the technique of comparative genomic hybridization and fluorescence in situ hybridization were performed to elucidate the histogenesis of non-gestational choriocarcinomas.^{14,18} Verbeek et al.¹⁸ found changes characteristic for colorectal adenocarcinomas, a loss of chromosomal regions 8p21-pter as well as 18q21-pter, and a gain of 5p and 20q, in both adenocarcinomatous and choriocarcinomatous tumor parts. This provides evidence for the common origin of both components.

Histopathologically, the diagnosis of choriocarcinoma is based on morphologic features with the finding of cytotrophoblasts and syncytiotrophoblasts with positive immunostaining to β -hCG.^{1,2,4} The absence of adenocarcinoma in some cases might be explained by the more aggressive choriocarcinomatous component that dedifferentiated it from adenocarcinoma.^{4,21} Immunohistochemical studies showed a significant proportion of morphologically typical adenocarcinoma of the large bowel disclosing positivity for hCG at

variable percentages and intensities.^{4,24} It seems that the appearance of hCG in colonic carcinomas is not an infrequent phenomenon. On the other hand, Lind and Haghighi¹² reported elevated serum carcinoembryonic antigen (CEA) levels and immunohistochemical positivity for CEA in neoplastic syncytiotrophoblasts.

ENC is an aggressive neoplasm that metastasizes through the lymph and blood streams. Metastatic disease is usually located in the lymph nodes, liver, lung, brain, and bones. As Table 1 shows, the prognosis associated with choriocarcinoma of the colon is very poor. Only two of the nine patients reported herein with the diagnosis of primary colonic adenocarcinoma with choriocarcinomatous differentiation underwent chemotherapy, and their survival was 16 and 60 months, respectively.⁵ The overall poor prognosis of patients with primary colonic choriocarcinoma may reflect the late diagnosis and the high volume of metastatic disease at the time of diagnosis. It seems that in cases of coexistent adenocarcinoma and choriocarcinoma, the latter appears to negatively affect the prognosis.²²

The response of colorectal choriocarcinoma to chemotherapy is much worse than the choriocarcinoma derived from germ cells for unknown reasons. It seems that the differences in chemosensitivity may be associated with the origin of carcinoma cells.^{5,22} The chemotherapy modality for primary extragenital choriocarcinoma has not been established as there is no standardized treatment. Recently, it was demonstrated that the collagen gel droplet-embedded culture drug sensitivity test (CD-DST), as a new in vitro anti-cancer test, offers precious therapeutic information for patients with colorectal cancer and pancreatic tumors.²² In their study, Maehira et al.²² suggest that CD-DST may provide—at least in part—therapeutic approaches for selecting suitable anti-tumor agents for patients with colorectal choriocarcinoma. Moreover, they performed a sigmoidectomy and administered FOLFOX and FOLFIRI plus bevacizumab, based on the results of the CD-DST test for colorectal choriocarcinoma, which resulted in repression of the tumor growth for 4 months.

In conclusion, we describe a patient with colonic adenocarcinoma with choriocarcinomatous dedifferentiation who presented with an unusual abscess formation. The report of this case is important to add data to the literature to help physicians keep in mind this challenging diagnosis and, eventually, to improve patient treatment and prognosis.

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The authors retain an informed consent signed by the patient's next-of-kin authorizing the publication of the data.

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Correspondence

Vasiliki Galani

Department of Anatomy - Histology and Embryology - School of Health Sciences - University of Ioannina

Dourouti, 45110 Ioannina, Greece

Phone: + 30 (2651) 007587

vgalani@uoi.gr