

Primary intestinal choriocarcinoma in a patient with long-standing Crohn's disease

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SUMMARY: Primary intestinal choriocarcinoma in a patient with long-standing Crohn's disease.

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Extra-gonadal choriocarcinoma is an extremely rare highly mali-

gnant neoplasm with a poor prognosis. In the gastrointestinal tract it usually arises in stomach, esophagus, bowel intestine and colon. Only few cases are pure and not associated with a classic adenocarcinoma. The correlation of Crohn's disease with choriocarcinoma is not reported. We describe a case of 47-year old man with primary choriocarcinoma of the colon in a previously documented Crohn's disease.

KEY WORDS: Choriocarcinoma - Adenocarcinoma - Extra-gonadal - Crohn's disease.

Introduction

Choriocarcinoma is a tumor that arises from a malignant transformation of trophoblastic cells in the female genital organs and from germ cells in the male gonads. A primary choriocarcinoma can occur in non gestational or extra-gonadal sites. In colon it is extremely rare and the prognosis is poor because of the low chemosensitivity. The association of choriocarcinoma with an inflammatory bowel disease is not reported to date.

Case report

We studied the case of a 47-year old male patient presenting with abdominal pain and melena. Twelve years before, he was affected by a perineal abscess, diagnosed as Crohn's disease on biopsy. Colonoscopy showed an ulcerated mass in the cecum that occupied the major part of the lumen. In the other sites, mucosa surface was hy-

peraeic. Subsequent full-body CT demonstrated the intestinal tumor and the presence of lung and brain lesions, likelihood of metastatic character. The patient underwent colectomy with lymph-nodes dissection. The tumor was located in the cecal colon, occupying almost all the diameter of the lumen and involving the ileo-cecal valve. The mucosal surface next to the tumor seemed to be edematous, relieved and blackish. The pathological findings showed that the tumor invaded the muscular layer and the pericolic fat with massive venular neoplastic embolism. It was described as a solid tumor, characterized by nests and trabeculae of epithelial and pleomorphic cells with numerous typical and atypical mitotic figures (Figure 1). The most part of the mass showed necrosis and hemorrhage and no glandular structures were identified. Histological examination of the healthy ileal mucosal specimens revealed edema, a mild polymorphic inflammatory infiltrate, glandular atrophy and architectural distortion, typical feature of Crohn's disease. Immunohistochemistry was positive for β -hCG (Figure 2) and also for CK-pool, CK-19, Vimentin and negative for Actin, CDX2, Desmin, CD34, CD68, CD45(LC), Melan-A (Mart1), S-100, DOG-1, CD117 and α -FP. All lymph-nodes were negative for neoplastic cells. To exclude the possibility of micrometastasis in the marginal zone, where atypical cells were found, immunohistochemical staining for CD68 and CK-pool was done, resulting negative. The histological and immunohistochemical features of this malignant neoplasm were com-

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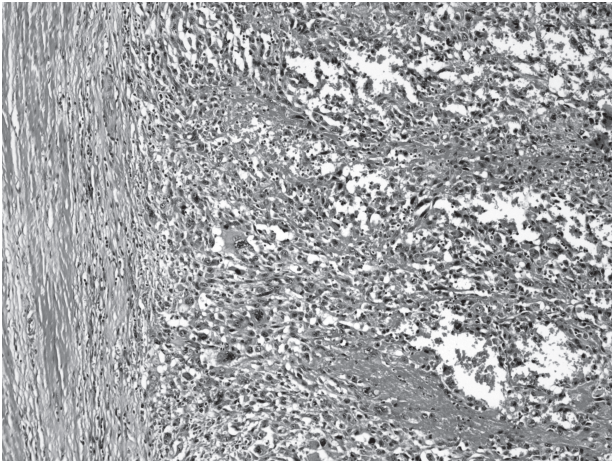


Figure 1 - Choriocarcinoma invaded the muscular layer with endovascular neoplastic embolism. Large and pleomorphic elements with numerous mitotic figures are evident (H&E, 200x).

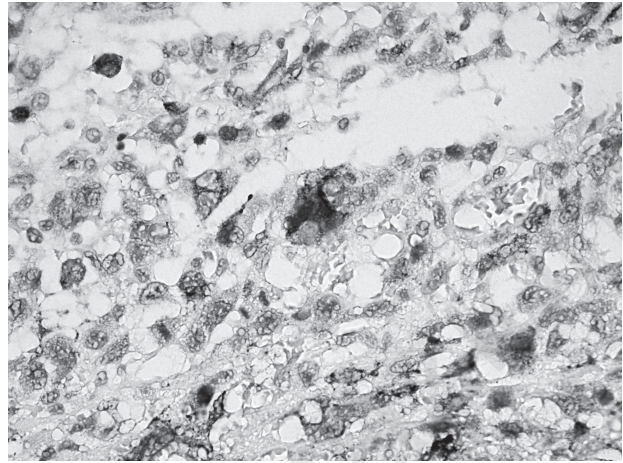


Figure 2 - Positive cytoplasmic immune-stain for β -HCG in neoplastic cells.

patible with a primary pure intestinal choriocarcinoma. No evidence of a testicular mass was found by radiological exams. Serum β -hCG levels haven't been tested because the patient had died in a little time.

Discussion and Conclusion

Intestinal choriocarcinoma is a rare tumor. Only 13 cases are described in literature (1). At the diagnosis, patients show widespread metastasis or develop them after a little time. A combination of radical surgery and adjuvant chemotherapy prolongs survival even if prognosis is poor. The pathogenesis of primary extra-gonadal choriocarcinoma is discussed. Three theories try to explain its origin. It could derive from primordial germinal cells migrated into these sites during the embryogenesis (2) or from a primary unknown lesion in the genital organs (3). Nevertheless, the most probable

hypothesis is that it arises from a dedifferentiation of a preexisting colic adenocarcinoma (4). This explanation is supported by the high percentage of cases in which choriocarcinoma is associated to a traditional adenocarcinoma. In the pure form choriocarcinoma could origin from a malignant transformation of totipotent cells in ectopic chorion or from an overgrowth of choriocarcinomatous clone in a poor differentiated adenocarcinoma. Only in a case choriocarcinoma is described in a woman with an ulcerative colitis but the relationship between these pathological conditions is not reported (5).

The report of this rare case of primary pure intestinal choriocarcinoma in a patient with an history of Crohn's disease is important to better know pathogenesis, diagnosis and management of similar cases and to improve knowledge about the possibility of a relationship between this rare malignant condition and a documented IBD.

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