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CASE REPORT

Testicular Choriocarcinoma Metastasizing to the Small Bowel Causing Intussusception: Case Report

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Introduction

Testicular cancers are divided into germ cell tumors (GCT) and non-germ cell tumors. Testicular germ cell tumors (TGCT) are the most frequent malignancy among young men although they are uncommon among general population [1]. There are recent evidences that the incidence of TCGT TGCTs is are increasing among Hispanic men with an expected increase over the next decade [2]. Germ cell tumors are either seminomas or non-seminomas (NSGCT) [3]. Testicular choriocarcinoma is a highly aggressive vascular germ cell tumor with trophoblastic differentiation [4]. It is usually detected as a component of the mixed malignant germ cell tumors. However, in a pure form, it is exceedingly rare and compromise only 0.2–0.6% of all testis tumors [5].

Testicular choriocarcinoma is characterized by its early hematogenous and lymphatic spread, with frequent metastasis to the central nervous system, lungs, bone, liver, and lymph nodes. It usually presents with symptoms related to metastases rather than a palpable mass in the testis [6]. Gastrointestinal tube involvement as a metastatic site for testicular choriocarcinoma is present in less than 5% of cases and its involvement worsens prognosis [7, 8]. The main symptoms of GIT metastasis are related to the presence of hemorrhage, or bowel obstruction or perforation. However, presenting as intussusception is exceedingly rare [9]. We are presenting a case of pure testicular choriocarcinoma with small intestinal metastasis intussusception.

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Clinical Summary

A 21-year-old Hispanic male presented to the emergency department in Texas Tech Health and Science center in El Paso, TX, with 12-h right lower quadrant abdominal pain. Pain was 10/10 in intensity, sharp in quality, with associated nausea and vomiting. But no fever (temperature 36.5 °C) melena nor hematochezia. By reviewing the past medical history of the patient, we knew that he was diagnosed by stage IV testicular pure choriocarcinoma with brain and lung metastasis 1 month before. He underwent orchiectomy, right parietal and left frontal craniotomy. He also had a family history (grandfather) of testicular cancer.

Physical examination showed blood pressure of 145/82, heart rate 96 bpm, and respiratory rate 18/min. His abdomen was soft, diffusely tender to palpation, with hypoactive bowel sounds, no organomegaly. The rest of physical examination was negative. His lab results showed pancytopenia due to chemotherapy. His white blood cell count was $0.8 \times 10^3/\mu l$; absolute neutrophil count was 0.59 cells/microliter. Hemoglobin was 6.0 g/dl. Platelet count was $31 \times 10^3/\mu l$. Computerized tomography (CT) scan showed intussusception.

Enterotomy surgical procedure involving the segment of ilium was done and sent to pathology. The resected segment was 18.0 cm in length, and diameter ranged from 6.0 to 8.5 cm. The overlying serosal surface was smooth and glistening. The resection margins were viable. At approximately 1.5 cm from the distal end, there was a firm hemorrhagic, necrotic polypoid-shaped lesion that protruded into the lumen and retracts the overlying serosal surface on the antimesenteric border. This lesion measured 2.3 × 1.8 cm

and 1.0 cm in thickness. Upon opening the lumen, it showed hemorrhagic fluid. The small intestinal mucosa adjacent to the polypoid mass was also necrotic with foci of intramucosal hemorrhage (Fig. 1).

Pathological Findings

The histological examination of the mass showed cytotrophoblasts and syncytiotrophoblasts consistent with metastatic choriocarcinoma (Fig. 2). The resection margins were negative for malignancy. Cells were positive for hCG immunohistochemical staining, while OCT-4, AFP, and CD30 immunhistochemical staining were negative (Fig. 3).

By reviewing the patient's records, his story started when he had a month of difficult voiding and pain in the right testicle that was not relieved by analgesics. The patient was diagnosed with stage IV testicular choriocarcinoma. Two weeks after orchiectomy, he started having paresthesia involving the left upper and lower extremities with numbness and severe intermittent headaches resulting in vomiting. Patient also reported at that time that he had difficulty in urinating with hematuria and blood in stool. On examination, there was weakness and paresthesia in left upper and lower limbs. He had slight nystagmus. CT scan of the brain showed right parietal lobe and left frontal lobe lesions. Chest CT scan showed metastasis to lungs. Patient underwent right parietal craniotomy and left frontal craniotomy. Lesions were diagnosed as metastatic choriocarcinomas with histopathology. Lesions were immunohistochemically positive for hCG. Two weeks after, the patient developed melena/hematochezia with some epigastric pain and loss of appetite. Upper endoscopy showed 4 mm duodenal polyp. Pathology showed normal mucosa with no evidence of celiac disease or malignancy. After 5 days, colonoscopy was done. But showed poor preparation and blood in lumen. So, capsule endoscopy was performed on the following day. It showed small bowel ulcers and blood in the small bowel and colon. The hCG level was 25, 506 IU/mL patient started after surgery on cisplatin, etoposide. Patient moved to tertiary care center.

Discussion

Testicular germ cell tumors represent 95% of malignant testicular tumors. Of them, 40–50% are seminomas. Non-seminomatous germ cell tumors include embryonal, teratomas, choriocarcinoma, or mixed testicular choriocarcinoma [10]. Testicular choriocarcinoma is a rare, highly aggressive vascular germ cell tumor. It is usually found as a component of malignant mixed germ cell tumors. While pure choriocarcinoma is less than 1% of testicular neoplasms. The microscopic picture of choriocarcinoma shows a biphasic pattern with mononucleated trophoblasts (cytotrophoblasts) and multinucleated syncytiotrophoblasts with hemorrhage, necrosis, and vascular invasion [11]. By using immunohistochemistry, syncytiotrophoblasts are positive for hCG, human placental lactogen, and inhibin. P63 immunoreactivity can be seen in cytotrophoblasts [4].

Vascular invasion, lymphatic invasion, and metastasis are very common and usually result in the primary symptoms. Usually, choriocarcinoma is associated with tumoral hemorrhage and patients may suffer from "choriocarcinoma syndrome" which is due to hemorrhage in the metastatic site [12]. Gastrointestinal tube involvement as a metastatic site present less than 5% of cases and its involvement worsens prognosis [7]. The stomach, usually the upper body, is the most common location, with a few case reports on the small intestine and colon [13–15]. Metastatic tumor implants reach the submucosa and stay there because they fail to pass through the capillary barrier. These submucosal tumors grow, resulting in insufficient blood supply to its central area, which leads to erosions and ulcerations. For this reason, the main morphologic endoscopic features of metastatic gastrointestinal lesions

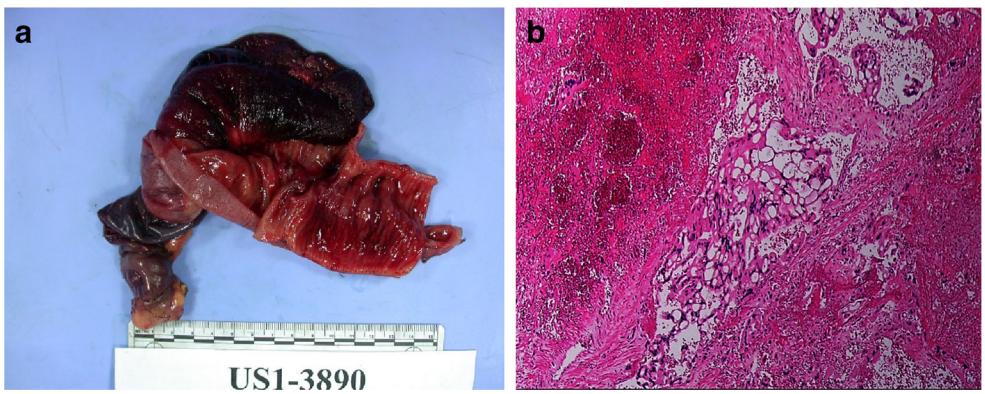

Fig. 1 **a** Small bowel lumen with an intussusception of adjacent small bowel. It has a dark, erythematous appearance, consistent with bowel necrosis. **b** Multinucleated, syncytiotrophoblasts in a background of necrosis and hemorrhage. No chorionic villi are identified (× 10)

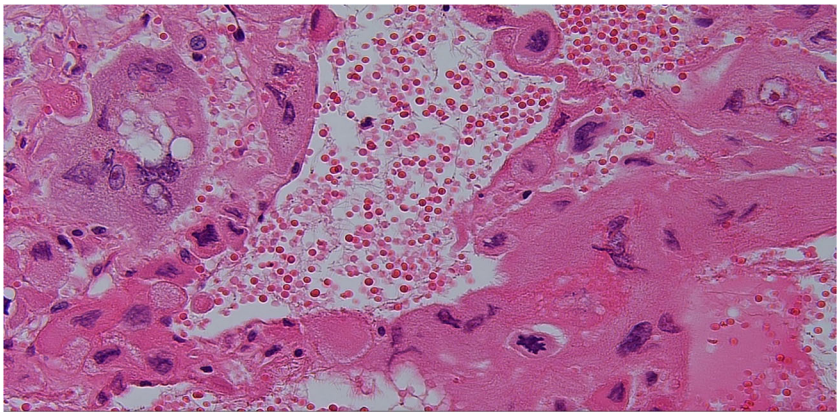

Fig. 2 Syncytiotrophoblasts seen in this metastatic choriocarcinoma (× 40)

are ulcerated submucosal and polypoid masses [16]. Extragenital primary choriocarcinoma in the small intestine is rare and usually presents as an ulcerated lesion. It should be included in the differential diagnosis of small intestinal neoplasm [17].

In literature, there are several cases of gestational and extra gestational female choriocarcinomas metastasizing to the GIT causing different symptoms ranging from hemorrhage, perforation, and rarely intussusception. For example, Nkanza and King [18] presented a case of choriocarcinoma presenting as an ileo-ileal intussusception in a 16-year-old girl. Ramessur et al. [19] presented a case of advanced gestational choriocarcinoma with small bowel metastatic involvement and intussusception. Popov et al. [20] presented a case of an 18-year-old nulliparous woman presenting with upper gastrointestinal bleeding and intussusception due to jejunal choriocarcinoma. Another case was of a 24-year-old woman with bowel obstruction secondary to intussusception caused by a metastatic choriocarcinoma polypoid mass [21]. While Ji et al. [22] presented a case of 33-year-old woman presented with gastrointestinal bleeding due to ileal invasion by metastatic tubal choriocarcinoma. In males, there are several case reports of testicular choriocarcinomas presenting with GIT metastasis and presenting with various symptoms like massive bleeding and perforation [21, 23, 24]. While by reviewing the literature using the words "testicular choriocarcinoma" and "intussusception." We found two cases in Spanish [21, 25] and two cases in English and they were caused by metastatic seminoma not choriocarcinoma. [26, 27]

Human chorionic gonadotropin is produced by the tumor cells. It is an important tool in both diagnosis and prognosis and also in predicting the response to treatment. It is high

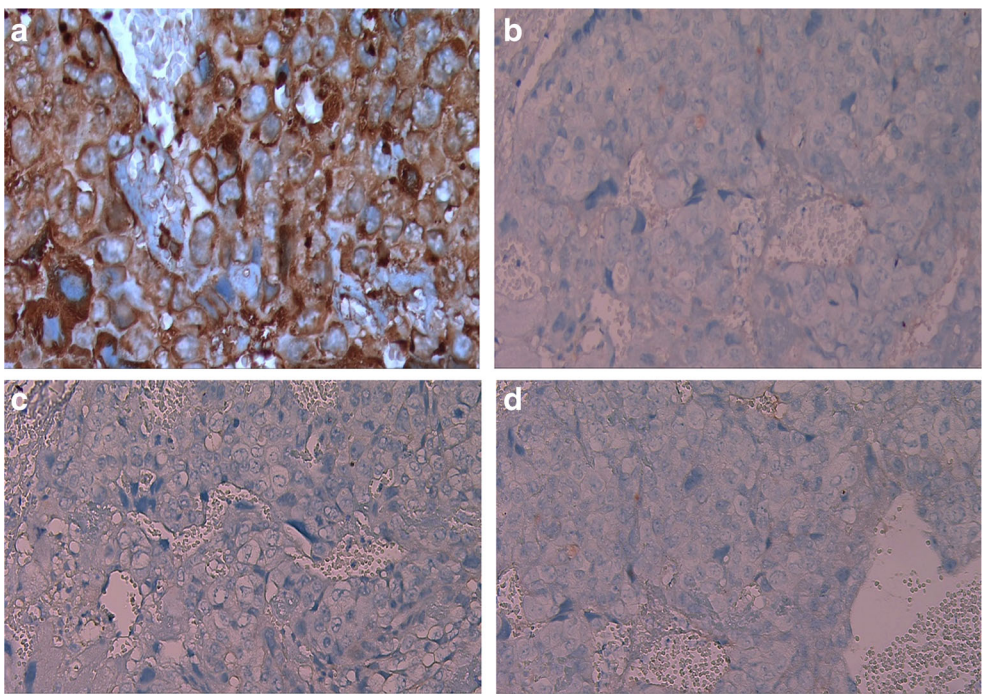

Fig. 3 **a** Positive immunohistochemical staining (IHC) for hCG. The rest of the pictures show negative IHC for **b** AFP, **c** OCT4, and **d** CD30

serologically and expressed immunohistochemically. An hCG level > 50,000 IU/L puts the patient in a poor prognostic group [28]. Prognosis of metastatic choriocarcinoma depends mainly on the chemo-refractory status, beta hCG serum concentration. However, the presence of GIT metastasis makes the prognosis worse [29].

In conclusion, testicular tumors should be excluded in any young man presenting with unexplained GIT symptoms in the form of bleeding, upper GIT obstruction, ulceration, or intussusception. On the other side, in known patients with testicular cancers, GIT involvement should be excluded especially if symptoms of obstruction or bleeding developed. These patients with GIT involvement will be in the poor prognosis category.

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Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflicts of interest.

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