CLINICAL CASE

Intestinal metastasis from mature testicular teratoma with transformation to choriocarcinoma


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KEYWORDS
Teratoma; Choriocarcinoma; Digestive tract bleeding; Metastasis; Mexico.

Abstract Testicular cancer represents 1% to 1.5% of neoplasias in men. The dissemination route of these tumors is through the lymphatic pathway to the retroperitoneal lymph nodes in 70% of the cases, with a lower frequency to the lungs, liver, and brain. Choriocarcinoma spreads by means of the bloodstream. The histologic malignant transformation of teratoma into rhabdomyosarcoma presents in up to 6% of cases. Metastatic gastrointestinal hemorrhage is rare. There are no previous reports on a change into choriocarcinoma or on a malignant transformation in the intestine, as a metastatic site.

The aim of this article was to describe a case of a germ cell tumor that transformed into choriocarcinoma in intestinal metastasis that presented as digestive tract bleeding.

A 23-year-old man was not able to eat orally, and presented with colic pain in the left iliac fossa radiating to the ipsilateral testis, accompanied by hematochezia. Digestive tract bleeding was attended to at the Emergency Service and no apparent hemorrhagic site was found. An increase in volume and induration of the right testis was reported and tumor markers were negative. Right radical orchiectomy was performed and mature teratoma was reported. Later, exploratory laparotomy was carried out due to the persistence of bleeding and it identified a bleeding tumor in the ileum. Thirty-six centimeters of the bowel were resected. Clinical stage III-C choriocarcinoma, T1 N1 M1b S1, was reported; adjuvant chemotherapy was indicated with 4 cycles of bleomycin, etoposide, and cisplatin (BeP) and 2 cycles of paclitaxel; at current follow-up, the patient presents no signs of disease recurrence or progression.

Mature teratoma metastases can transform into different histologies. Metastasis to the small bowel from germ cell testicular tumors is rare and manifests as bowel obstruction and digestive tract bleeding. Transformation into choriocarcinoma has not been reported.
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PALABRAS CLAVE
Teratoma; Coriocarcinoma; Sangrado tubo digestivo; Metástasis; México.

Introduction

Testicular cancer represents 1% to 1.5% of the tumors in men and 5% of urologic tumors in general. In the Western countries, 3 to 10 new cases per 100,000 men present every year. The predominant histologic type (90%-95%) is germ cell tumor. These are more common in men from 15 to 35 years of age. The dissemination route is through the lymphatic system to the retroperitoneal lymph nodes in 70% of these tumors, and at lower percentages, to the lungs, liver, and brain. Choriocarcinoma spreads by means of the bloodstream. Metastases of germ cell testicular tumors to the gastrointestinal tract are rare. Those patients that present with intestinal metastases can manifest the disease with symptoms of bowel obstruction and digestive tract bleeding at the time of diagnosis and during treatment. Treatment is surgical resection of the metastatic site and chemotherapy in the majority of cases, increasing morbidity and mortality. Up to 6% of teratomas have a malignant transformation to rhabdomyosarcoma; other transformation possibilities are adenocarcinoma, osteosarcoma, neurosarcoma, and myxoid sarcoma. Likewise, the most frequent malignant transformation sites correspond to the retroperitoneum, mediastinum, lung, pleura, and testis. There are no previous reports on the conversion to choriocarcinoma or on malignant transformation in the intestine as the metastatic site.

The aim of the present article was to describe a case of testicular germ cell tumor that transformed to choriocarcinoma with metastasis to the intestine, manifesting with lower digestive tract bleeding.

Case presentation

A 23-year-old male had disease onset one week prior with an inability to eat by mouth, colicky pain in the left iliac fossa of 7/10 intensity accompanied by nausea and vomiting, pain radiating to the ipsilateral testis, and hematochezia on numerous occasions. The Urology Service
stabilized the digestive tract bleeding and then performed endoscopy and colonoscopy, without finding the apparent site of the bleeding. An increase in volume and induration of the right testis were documented and an ultrasound study showed a testicular mass with hypoechoic, heterogeneous images affecting the testicular parenchyma; they were non-cystic and with no vascularity (fig. 1). Tumor markers were negative, alpha-fetoprotein was 3.17 ng/mL, lactate dehydrogenase 185 U/L, and human chorionic gonadotropin hormone 2.8 ng/mL. The chest x-ray showed no metastasis. Right radical orchiectomy was performed and the histopathologic report stated a 5.3 cm mature teratoma with negative surgical margins, no lymphovascular invasion, and stage T1 N1 M1b S1. In the mediate postoperative period, the patient presented with bowel obstruction and had signs of active digestive tract bleeding. Abdominal contrast-enhanced computerized axial tomography (CAT) scan identified 2 non-liquid, free, intraluminal ovoid images in the ileum (fig. 2) and a retroperitoneal lymph node of approximately 1 cm (fig. 3). Exploratory laparotomy was done at the site of the bleeding tumor in the ileum; it was managed with resection of 36 cm of the intestine and end-to-end anastomosis. The histopathologic study reported choriocarcinoma in the ileum with positive surgical margins. Thus it was classified as having a clinical stage of III-C, T1 N1 M1b S1. Adjuvant chemotherapy was begun with 4 cycles of bleomycin, etoposide, and cisplatin (BEP) and 2 cycles of paclitaxel, due to the persistence of retroperitoneal lymph nodes (table 1). The patient is currently in follow-up with no evidence of disease recurrence or progression (table 1) and the control CAT scan showed retroperitoneal lymph nodes under 1 cm.

Discussion

Mature teratoma metastases can transform into different histologies, and some that have been reported are adenocarcinoma, rhabdomyosarcoma, or neuroectodermal tumor.

Metastasis to the small bowel from testicular germ cell tumors is rare and no percentage has yet been reported. Among its possible manifestations are bowel obstruction and digestive tract bleeding, requiring surgical management the majority of the time. The most frequent histology in these cases is mixed germ cell tumor. However, transformation to choriocarcinoma has not been reported. This is why early and opportune diagnosis and treatment are necessary in patients with germ cell tumors with intestinal metastasis. Thus, patients with a known diagnosis of testicular cancer, especially those with histologic characteristics of choriocarcinoma and involving the digestive tract, are regarded as advanced disease, since reported cases have received chemotherapy for presenting with metastases in other sites at the same time. Some conventional imaging studies do not show occult metastatic masses, and so symptomatology such as nausea, vomiting, abdominal pain, or digestive tract bleeding should make us think of the possibility of intestinal metastases from testicular cancer. Patient outcome for gastrointestinal metastases has been reported as poor.
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Metastatic disease is detectable at the time of presentation in 30% of the patients with seminoma and in 50% of those with non-seminomatous germ cell tumors. Generally, choriocarcinoma has rapid proliferation and excessive vascularity. Choriocarcinomas with these characteristics have a tendency for ulceration, necrosis, or bleeding. The majority of metastases to the gastrointestinal tract occur as disease at other sites that has already progressed. There are 2 different methods by which testicular germ cell tumors involve the gastrointestinal tract: a) direct infiltration by the tumor due to retroperitoneal lymph node involvement and b) as a result of hematogenic dissemination. Direct infiltration generally occurs in the small bowel, especially in the duodenum (in approximately 95%). Symptoms of anemia secondary to gastrointestinal bleeding can appear and blood loss can be occult or massive. In a study by Johnson, of the 16 patients with choriocarcinoma, 7 presented with hemorrhagic complications, 4 of whom died from those complications. The approach to digestive tract bleeding is similar to that used for upper or lower digestive tract bleeding; it is generally by endoscopy or colonoscopy.

In our case, as in others, the pathology was not found at a site that can be reached through endoscopy, since it originated in the ileum. In such cases an angio gram of the superior mesenteric artery with marked erythrocytes is indicated. Two mechanisms by which the malignant transformation of a teratoma is carried out are: 1) malignant differentiation of somatic type totipotential embryonal carcinoma cells or 2) malignant transformation of mature teratoma elements. Some teratomas are malignant due to post-teratoma malignant transformation; these tumors usually occur in the testis and exhibit a spectrum of biologic behavior. High-grade examples include dermoid cyst with squamous cell carcinoma or various sarcomas or melanomas, and primitive neuroectodermal tumor.

Conclusions

Testicular tumor metastases to the intestine are rare and can manifest as symptoms of digestive tract bleeding, increasing morbidity and mortality. This is the first case in the national medical literature reporting the transformation of teratoma to choriocarcinoma in the intestine as the metastatic site.

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Conflict of interest

The authors declare that there is no conflict of interest.

References