Spontaneous rupture of a retroperitoneal lymph node metastasis causing massive hemorrhage in a patient with advanced mixed germ cell tumor

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ABSTRACT

Retroperitoneal hemorrhage is a rare and life-threatening clinical presentation that may occur in patients with advanced germ cell tumors, typically after chemotherapy. Rapid clinical suspicion is crucial to offer the best treatment. Surgery is usually recommended but dismal complications may occur. We report on a man with metastatic testicular cancer presenting with massive spontaneous retroperitoneal hemorrhage that had not received previous systemic treatment and was managed without surgical treatment, with good clinical outcome.

1. Introduction

Germ cell tumors (GCTs) are a common malignancy among men aged 15–35 years that characteristically metastasize to the retroperitoneal lymph nodes, lungs, liver, bones or brain. There are few reports in the literature about metastatic germ cell tumors causing massive hemorrhage. This rare and threatening situation may occur in advanced germ cell tumors with high-volume choriocarcinoma elements, typically after chemotherapy.

We report an unusual presentation of spontaneous rupture of retroperitoneal metastases of a stage III non-seminoma germ cell tumor, in which conservative management had a favorable outcome.

2. Case report

A 24-year-old male patient presented to the emergency department reporting right flank pain for about a week before admission, with irradiation to the right scrotum. The patient was otherwise healthy. On physical examination, a mass was felt in the right hypochondrium, painless on palpation with approximately 5 cm in diameter. The right testicle was enlarged with a 2 cm nodule in the upper pole. The left testicle had no identifiable alterations.

An ultrasound examination of the scrotum revealed a mixed solid/cystic hypoechoic nodule with regular contours in the upper pole of the right testicle, measuring 2.1 × 1.6 × 1.2 cm, with peripheral and central vascularization at Doppler. Computed tomography (CT) showed the presence of multiples nodes to both lungs as well as a solid and heterogeneous retroperitoneal mass compatible with metastasis measuring 7.6 × 6.6 × 10cm. (Fig. 1). Laboratory tests revealed elevated DHL of 588 U/L, beta-HCG of 227mUI/mL, and alpha-fetoprotein of 1670 ng/mL.

The patient underwent an uneventful left radical orchiectomy. Pathology report showed a non-seminomatous tumor with endodermic (55%) and teratoma (45%) components, with free surgical margins and no rete testis involvement.

Six days after the orchidectomy, the patient was readmitted to the hospital with abdominal pain of sudden onset associated with nausea and vomiting. Vital signs showed hypotension and tachycardia. Upon physical examination, a tense abdomen with diffuse tenderness and an apparent enlargement of the abdominal mass were identified. Laboratory tests showed a significant drop in hemoglobin levels from a previous value of 17.1 g/dL to 8.2 ng/dL at readmission. A CT of the abdomen and pelvis showed an increase in the dimensions of the interaortocaval mass, now measuring about 15 × 10cm suggesting hematoma and a moderate amount of free fluid in the abdominal cavity. (Fig. 2).

Patient was admitted to the intensive care unit for hemodynamic stabilization. The use of vasoactive drugs was not required. Ultimately, the patient did not require transfusion and was discharged after 5 days of intensive care with hemoglobin levels at 8.0 g/dL. No surgical procedures were performed.

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Medical oncology opted to initiate the first cycle of systemic treatment a few days after the intensive care discharge, with the patient still admitted to the hospital, with etoposide and carboplatin. After one week, clinical fully response was observed and stabilization of hematometric levels was maintained. Without complaints, discharge occurred after fifteen days of hospitalization. The patient showed no adverse effects. Two months later, a re-staging workup showed significant reduction of the retroperitoneal mass, with no new episodes of bleeding and later, a favorable response to treatment (Fig. 3).

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3. Discussion

Hemorrhage caused by spontaneous rupture of retroperitoneal metastasis is a rare complication of testicular tumors, seldom described in literature. Most cases were associated with choriocarcinoma, which is defined as “choriocarcinoma syndrome”. Some autopsy series have reported that up to 44% of patient deaths due to testicular choriocarcinoma are caused by bleeding events. Initiation of chemotherapy may represent the most significant risk factor for bleeding. Surprisingly, in our case, the patient had not yet received systemic treatment and the tumor had no choriocarcinoma component.

A surgical approach was recommended in the few similar cases reported in the literature. Komori et al. reported on the surgical approach of retroperitoneal lymph node hemorrhage in a patient with a metastatic mixed germ cell tumor. Intraoperatively, about 2 L of blood was lost. The patient survived the surgical procedure, but succumbed to complications one month later. Moore et al. reported on a case of spontaneous rupture of a retroperitoneal mass in which laparotomy was performed and blood loss was approximately 10 L.

One must be aware that causes of retroperitoneal bleeding may also include the possibility of lesions to the great vessels during chemotherapy. Mego et al. describes a patient in which an aortic rupture occurred one day after systemic PEB treatment was administered. CT scan showed a dorsal aortic wall rupture with active extravasation and irregular pseudoaneurysmatic dilation of the aorta. Overall, the morbidity of the surgical exploration is considerable, and urologists should be aware that dismal surgical complications may arise when determining the best course of action for such cases.

Based on our experience, conservative treatment should be considered initially in the management of similar cases. However, hemodynamic instability requiring the use of vasoactive drugs and multiple blood transfusions, surgical approach should be discussed. As it represents a rare and potentially lethal situation, and there are no specific guidelines, the clinical management must be individualized.

4. Conclusion

Spontaneous retroperitoneal hemorrhage is a rare complication of metastatic testicular tumor that requires expedient care. Special attention should be given to patients with choriocarcinoma elements in the pathological specimen undergoing chemotherapy. Surgical approach has a high morbidity. Conservative treatment may be an option for selected patients. Sound clinical judgment is imperative for a good outcome.

References

1. Schmoll HJ, Souchon R, Krege S, et al. European consensus on diagnosis and treatment of germ cell cancer: a report of the European Germ cell Cancer Consensus Group. Ann Oncol. 2004;15:1377–1399.
2. Car, Reekers JA, Westermann AM, Van der Velden J. Embolization for hemorrhage of liver metastases from choriocarcinoma. Gyneco Oncol. 2005;98:506–509.
3. Komori K, Takahari D, Kimura K, et al. Recovery from choriocarcinoma syndrome associated with a metastatic extragonadal germ cell tumor hemorrhage. Case Rep Gastroenterol. 2016;10:198–203. https://doi.org/10.1159/000446017.

4. Moore K, Imbeault A, Roy G, Bolduc S. Massive hemorrhage from spontaneous rupture of a retroperitoneal lymph node in patient with metastatic mixed germ cell tumor. Urology. 2010 Jul;76(1):159–161. https://doi.org/10.1016/j.urology.2009.10.001.

5. Hulova S, Azizi R, Volev I, et al. Successful emergency endovascular aortic repair for intratumoral hemorrhage in extensive retroperitoneal mass of testicular origin. BMC Surg. 2020 Nov 7;20(1):272. https://doi.org/10.1186/s12893-020-00933-2.