Acute pulmonary hemorrhage associated with metastatic testicular choriocarcinoma in a 46-year-old incarcerated male

Hali Pearce, Daniel C Edwards1, Jason A Levy1, Brian H McGreene, Lynn Mackovick1, Matthew Brennan2, Mary McHugh2, Beth Mapow1, Francis J Schanne1, Laurence Belkoff1

Alabama College of Osteopathic Medicine, Dothan, Alabama, 1Department of Urology, Hahnemann University Hospital, 2Department of Urology, Einstein Medical Center, Philadelphia, Pennsylvania, USA

Abstract

Pure testicular choriocarcinoma is a rare histological subtype of germ cell tumor (GCT) and typically presents with distant metastases and aggressive features leading to a generally poor prognosis. Unique to choriocarcinoma among GCT histological subtypes is the propensity of spontaneous hemorrhage into metastatic lesions. We report a case of pure testicular choriocarcinoma in a 46-year-old male with postoperative acute pulmonary hemorrhage secondary to tumor invasion of the lungs, and the subsequent management of his disease with a discussion of relevant literature.

Keywords: Choriocarcinoma, germ cell tumor, pulmonary hemorrhage, testicular cancer

INTRODUCTION

Primary testicular tumors are the most common solid malignant tumor in young men in the United States.[1] Germ cell tumors (GCTs) are the most common subtype of testicular malignancies. While normally identified as components of a mixed GCT, choriocarcinoma as an isolated histology comprises only 1% of GCT.[2] Choriocarcinoma syndrome (CS), a rare and lethal complication, presents as hemorrhage from metastatic sites and can manifest at any time during treatment, but frequently after the initiation of chemotherapy.[3]

CASE REPORT

A 46-year-old incarcerated male presented to the emergency department with the primary complaint of nausea, vomiting, and weight loss. His medical history was significant for a diagnosis of schizophrenia and history of right-sided cryptorchidism, status postinguinal orchiopexy at the age of 13 years. He had a 20 pack-year smoking history. He denied hematuria, dysuria, fevers, or chills. Overall, he was a poor historian of questionable mental status. On physical examination, it was noted that he had a large, firm, immobile right-sided scrotal mass that was tender to examination. The patient reported that this mass had been present and enlarging over the previous several months.

Initial laboratory values revealed a mild leukocytosis (13.0 × 10³), anemia (9.1 g/dL), normal alpha -fetoprotein (AFP) (1.0 ng/mL), elevated lactate dehydrogenase (LDH) (715 U/L), and elevated beta-human chorionic gonadotropin (b-HCG) (136,017 mIU/mL). Scrotal...
ultrasound revealed a 12.5 cm heterogeneous scrotal mass with central necrosis [Figure 1a]. Chest X-ray revealed diffuse pulmonary metastases [Figure 1b]. Magnetic resonance imaging (MRI) of the brain was ultimately performed for questionable altered mental status and revealed a small enhancing lesion [Figure 1c].

He underwent open right radical inguinal orchiectomy, which was complicated by his previous inguinal surgery and dense adherence to the right hemiscrotum. Gross examination demonstrated a large, centrally necrotic lesion [Figure 2a and b]. Final pathology revealed pure choriocarcinoma with a positive inferior scrotal margin. Noted on histopathology were a complete replacement of parenchyma with tumor and epididymal invasion [Figure 2c], multinucleated syncytiotrophoblasts [Figure 2d], and tumor emboli [Figure 2e]. Postoperatively, the patient developed respiratory distress requiring non-rebreather mask oxygenation and was weaned to nasal cannula over the following days. This was initially attributed to history of smoking and the large pulmonary metastatic burden. On postoperative day (POD) #2, he was started on etoposide, ifosfamide, and cisplatin chemotherapy. However, he remained tachycardic with increasingly worse oxygen saturation. CT angiogram was performed to evaluate for pulmonary embolism (PE), which demonstrated a singular, small left lower lobe PE, determined to be an unlikely contributor to his profound hypoxemia [Figure 3]. On duplex ultrasound, no lower extremity venous thrombosis was identified. Nevertheless, heparin anticoagulation was initiated. The same day, he developed severe hemoptysis and increased oxygen requirements. Anticoagulation was held until hemoptysis resolved, at which point, he was transitioned to apixaban. Ultimately, during a subsequent admission, he was transitioned to an inferior vena cava filter secondary to gastrointestinal bleeding, suspected to be from potential gastrointestinal metastases.

bHCG levels were followed over the subsequent months. On POD #14, bHCG was 34,238 mIU/mL, 1590 mIU/mL at 1 month, and 74 mIU/mL at 3 months. The patient continued on the previous chemotherapy regimen and is currently on his fourth cycle with no further hemorrhagic events.

**DISCUSSION AND MANAGEMENT**

The incidence of testicular cancer has increased during the last century, but testicular tumors are still a rare occurrence. Nevertheless, primary testicular tumors are the most common solid malignant tumor in young men in the US.[1] According to the National Cancer Institute's Surveillance, Epidemiology, and End Results Program, testicular cancer has an annual incidence of 5.7 cases per 100,000 people. In 2014, there was an estimated prevalence...
Testicular choriocarcinoma is classified as a subtype of nonseminomatous GCT (NSGCT). Choriocarcinoma is rarely identified in its pure form and is commonly found as a component of a mixed GCT. Pure testicular choriocarcinomas make up 1% of GCT cases. Testicular choriocarcinoma is significantly more aggressive and chemo-refractory than other subtypes of testicular cancer, with a 5-year survival rate <80%. Testicular cancer primarily affects men between the ages of 20 and 34 years; however, the majority of choriocarcinoma cases affect men between the ages of 25 and 30 years. Choriocarcinoma has a high likelihood of early metastasis, primarily to the lungs, liver, and brain. The lungs are involved in almost 100% of metastatic cases.

Typical presenting features of a choriocarcinoma include a man in his third to the fourth decade with a testicular mass and elevated serum bHCG. Signs and symptoms may include bilateral tender gynecomastia, hematogenous metastases (most commonly to the lungs bilaterally, presenting with a “snowstorm” appearance on chest X-ray), hyperthyroidism and/or hemoptysis.

A rare and lethal complication of the disease, CS is defined as hemorrhage from metastatic sites. If the metastases are in the lung, pulmonary hemorrhage, and acute pulmonary decompensation can result due to tumor invasion into pulmonary vasculature. CS is more likely with higher levels of bHCG. The most common presentation of CS is pulmonary hemorrhage, but hemorrhage can develop in the liver, brain, and small bowel as well. Hemorrhage can occur immediately following chemotherapy, or present as the initial primary complaint.

The current patient had a history of ipsilateral cryptorchidism status postorchidopexy at age thirteen. Cryptorchidism is considered a risk factor for infertility and testicular GCTs. Cryptorchidism is a common diagnosis in newborn male infants, with around 2%-4% diagnosed with unilateral or bilateral cryptorchidism. About 10% of all cases of testicular GCTs occur in men with a history of cryptorchidism, and the risk of developing testicular cancer due to cryptorchidism is increased 5–10 times than that of the general male population. The current guidelines for the management of congenital cryptorchidism is orchidopexy between 6 and 18 months of age if there is not spontaneous resolution.

When presented with a suspicious testicular mass, workup includes a thorough history and physical, AFP, bHCG, and LDH levels, a complete chemistry profile, and testicular ultrasound. Postdiagnostic workup of an NSGCT includes chest/abdominal/pelvic computed tomography, repeat bHCG, LDH, and AFP levels, and brain MRI if indicated. NSGCTs of all stages can be treated with primary chemotherapy using tumor markers to follow response. The standard chemotherapy regimen for poor-risk patients is four cycles of BEP (bleomycin, etoposide, and cisplatin); however, four cycles of VIP (etoposide, cisplatin, and ifosfamide) can be used in patients who cannot tolerate bleomycin due to pulmonary toxicity. If bHCG levels fail to normalize, additional cycles should be implemented. If bHCG levels plateau after several rounds of chemotherapy, the patient has refractory disease and more aggressive treatment should be considered including high-dose chemotherapy with stem-cell rescue. bHCG levels <10 mIU/mL indicate an acceptable response to treatment.

CONCLUSION

Pure testicular choriocarcinomas are a rare occurrence and can be complicated by the development of CS. Metastatic choriocarcinoma must be treated with a chemotherapeutic regimen, and bHCG should be followed to ensure response to treatment in addition to imaging parameters.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

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