Testicular Choriocarcinoma Presenting with Massive Hemoptysis and Endobronchial Metastases

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Abstract

Introduction: Pure testicular choriocarcinoma is a rare type of non-seminomatous germ cell tumors that affects young males, occurring in less than 1-5% of testicular cancers. It is the most aggressive form of germ cell tumors and tends to metastasize hematogenously to the lungs, liver, brain and other visceral organs with the testicular primary tumor can be small or burnt out. It responds to chemotherapy but carries high mortality rate.

Presentation of case: we report a case of 20 year-old otherwise healthy male who presented to the emergency department with massive life-threatening hemoptysis secondary to metastatic pure testicular choriocarcinoma, including endobronchial metastases. He responded remarkably to cisplatin-based chemotherapy followed by radical orchiectomy resulting in ongoing remission.

Conclusion: This case illustrates the clinical course of a young male with advanced primary testicular choriocarcinoma presenting with massive hemoptysis secondary to lung metastasis including endobronchial metastasis, which responded to cisplatin-based chemotherapy. Early recognition, joint management by intensivists, oncologists and urologists, and prompt initiation of cisplatin-based chemotherapy followed by orchiectomy forms the cornerstone of management.

Keywords: testicular germ cell tumors; testicular choriocarcinoma; pulmonary hemorrhage; massive hemoptysis; choriocarcinoma syndrome; endobronchial metastases

Academic Editor: Xiaoning Peng, Hunan Normal University School of Medicine, China

Received: April 27, 2015; Accepted: May 29, 2015; Published: June 7, 2015

Competing Interests: The authors have declared that no competing interests exist.

Consent: We confirm that the patient has given the informed consent for the case report to be published.

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Introduction

Testicular germ cell tumors (GCTs) represent 95% of primary testicular neoplasia in males [1-3]. Pure choriocarcinoma is the least common type of non-seminoma germ cell tumors (NSGCTs) occurring in less than 1-5% of cases [1, 3, 4]. However, it is the most aggressive variant and is characterized serologically by the production of large amounts of beta-Human Chorionic Gonadotropin (β-HCG). Although choriocarcinoma may metastasize to the lungs, endobronchial involvement is rare with only one case reported in the literature [5]. The choriocarcinoma syndrome, characterized by bleeding at the metastatic sites, is a rare and serious complication of choriocarcinoma tumor and is associated with poor prognosis [1, 6]. We report a case of a young male with pure testicular choriocarcinoma who presented with life-threatening hemoptysis secondary to choriocarcinoma syndrome and endobronchial metastasis, which responded to cisplatin-based chemotherapy followed by radical orchiectomy with ongoing remission.

Case presentation

A 20 year-old male otherwise healthy and on no medications, presented to the emergency department of a community hospital with a two week history of progressive exertional breathlessness, streaky hemoptysis and feeling unwell. On initial assessment, his blood pressure was 150/96 but he was tachycardiac (125/min), tachypneic (28/min) and hypoxic (SaO₂ 89% on room air). A chest radiograph showed right paratracheal and subcarinal lymphadenopathy, bilateral non-calcified pulmonary nodules and ill-defined heterogeneous air-space disease in the right lower lobe (Figure 1).

Figure 1 Initial Chest radiograph showing bilateral pulmonary nodules, right paratracheal and subcarinal lymphadenopathy (asterisks) and ill-defined heterogeneous air-space disease within the right lower lobe segments. Three discrete round non-calcified pulmonary nodules, 9 mm and 8 mm in the right upper lobe and 13 mm in the lingula.
Shortly after presentation, he developed massive hemoptysis and hemodynamic collapse requiring mechanical ventilation and resuscitation with intravenous fluids. An emergent bronchoscopy was performed. Brisk bleeding was noted from the right mainstem bronchus. The patient was placed in the right lateral decubitus position to protect the left lung; a right-sided bronchial blocker was successfully placed. Following stabilization, he was transferred to our tertiary centre for ongoing management in the intensive care unit (ICU).

On arrival to the tertiary centre, bronchoscopy was repeated. A large blood clot had formed at the orifice to the right mainstem bronchus (Figure 2a); the bronchial blocker remained well positioned in the right mainstem bronchus. The blocker was deflated, and no further active bleeding was observed. The bronchoscope was then gently maneuvered past the clot, and an abnormal, irregular, friable mucosa was visualized in the right mainstem bronchus (Figure 2b) that had the macroscopic appearance of endobronchial tumor.

**Figures 2** Bronchoscopy performed on presentation: 2a. Initial Bronchoscopy revealed significant narrowing of the right mainstem bronchus and a large blood clot at the orifice. A right bronchial blocker was also shown. 2b. An abnormal, irregular, friable mucosa was visualized in the right mainstem bronchus past the blood clot characteristic of an endobronchial tumour.

Physical examination was remarkable for a palpable right posterior cervical lymphadenopathy. Auscultation of the chest demonstrated decreased breath sounds on the right side with coarse crackles. Scrotal examination revealed bilateral small testicles (~ 3cm) and varicoceles, but no discrete masses were appreciated. There was no gynecomastia.

Computed tomography scan of the chest and abdomen showed extensive airspace disease in the right lung compatible with blood, multiple bilateral pulmonary nodules and extensive mediastinal and retroperitoneal lymphadenopathy (Figures 3a, 3b).
Figure 3 Computed Tomography scan of the chest: 3a. Coronal section of the chest showing extensive airspace disease predominantly in the right lung field compatible with blood. Endotracheal tube (ETT) in the proximal left mainstem bronchus (thick black arrow). There is mediastinal subcarinal lymphadenopathy (asterisk). There is also bilateral pneumothoraces and two pulmonary nodules in the left lung apex and left lower lobe (white arrows). 3b. Axial section of the chest showing severely narrowed right mainstem bronchus (thin black arrow). ETT is shown in the left mainstem bronchus (thick black arrow). There is right paratracheal lymphadenopathy (asterisk). There is also extensive airspace disease predominantly in the right lung field, bilateral pneumothoraces and pneumomediastinum.

Laboratory investigations showed very high levels of β-HCG (61928 IU/L; normal 0-2.5 IU/L). Serum lactate dehydrogenase (LDH) was mildly elevated (381 U/L; normal range 94-250 U/L), but alpha-fetoprotein (AFP) was within the normal range (5.4 ug/L; normal range 0-10 ug/L). Other laboratory investigations were unremarkable.

Testicular ultrasound showed a suspicious ill-defined hypoechoic lesion in upper to mid pole of the left testicle with increased vascularity and multiple coarse calcifications suspicious for testicular carcinoma (Figure 4). It also showed bilateral varicocele suggestive of retroperitoneal lymphadenopathy.

Figure 4 Ultrasound of the left testicle contains multiple coarse calcifications with a subtle ill-defined hypoechoic area in the upper to mid pole (arrows); appearance suspicious of testicular carcinoma.
Ultrasound-guided biopsy of the right cervical lymph node was performed. Microscopic examination revealed extensively necrotic, high-grade malignant neoplasm consistent with metastatic choriocarcinoma.

In consultation with medical oncology and urology, chemotherapy with cisplatin and etoposide (EP) was administered emergently within 72 hours after presentation. Bleomycin was omitted because of risks of pulmonary toxicity given his extensive lung involvement with respiratory failure. Following two cycles of EP chemotherapy, he showed a remarkable response with clinical, radiographic and bronchoscopic (Figure 5) evidence of disease regression.

Biochemically, his β-HCG level decreased to 34 IU/L. Although he required tracheostomy, he was successfully weaned from mechanical ventilation after 18 days. The tracheostomy was subsequently removed and the patient was discharged home one month later. As an outpatient, he received additional two cycles of etoposide, ifosfamide and cisplatin (VIP) chemotherapy, and underwent left radical orchiectomy. Pathology identified scarring and calcification within the left testicle with no residual malignancy. He remains well with ongoing biochemical evidence of disease remission (β-HCG <1.0 IU/L).

**Discussion**

Massive hemoptysis is a medical emergency and is associated with a high mortality. The most common etiologies are pulmonary malignancies and chronic inflammatory conditions such as bronchiectasis [7]. Death usually results from asphyxia secondary to flooding of the airways and alveoli with blood. The first priorities in management are to secure the airways, optimize oxygenation and achieve hemodynamic stability. Early bronchoscopy is crucial for localization and control of the bleeding culprit [7].

Testicular neoplasms compromise the most common solid malignancy in men between 15-35 years of age, but they only represent 1% of all solid tumors in males [1, 2]. More than 90% of primary testicular neoplasms are GCTs. Pure choriocarcinoma is rare, accounting less than 1-5% of testicular GCTs [1-3]. It is the most aggressive form and is characterized by rapid proliferation, high vascularity and tendency to invade the blood vessels leading to early hematogeneous dissemination. Yet, despite their aggressive nature, testicular physical examination may not demonstrate a clear palpable testicular mass [8].
Metastatic choriocarcinoma can be complicated by a rare, life threatening complication, known as the choriocarcinoma syndrome \[1, 2, 4, 6, \text{ and } 8\].

Logothetis first described the choriocarcinoma syndrome \[1, 6\]. It is a distinct clinical presentation of advanced GCTs with high-volume choriocarcinoma elements. It is associated with markedly elevated $\beta$-HCG levels. It is a life threatening condition, associated with poor prognosis and requires early recognition, emergent stabilization and administration of cisplatin-based chemotherapy \[1, 6\]. The hallmark of this syndrome is bleeding at the metastatic sites. It can occur in two clinical settings; more commonly few hours after the initiation of chemotherapy, which represent necrosis in this highly vascular tumor that is responsive to cytotoxic medications \[2, 8\]. It also might occur spontaneously at the metastatic foci as a result of invasion of the blood vessels \[2, 4, 6, 8, 9\]. Pulmonary hemorrhage secondary to lung metastasis is the most common presentation. However, these hemorrhagic manifestations can occur anywhere at the site of metastasis such as intracranial bleeding secondary to brain metastasis and hemoperitoneum attributable to liver or peritoneal metastasis \[2\].

Endobronchial involvement from choriocarcinoma is rare, and there is only one other reported case in the literature \[5\]. Given the highly vascular nature of choriocarcinoma, it is not surprising that massive hemoptysis may occur as a manifestation of endobronchial metastases.

**Conclusion**

This case illustrates the clinical course of advanced pure testicular choriocarcinoma presenting with massive hemoptysis secondary to lung metastasis. Also, It is the second worldwide reported case in the literature of choriocarcinoma presenting as an endobronchial tumor. Massive hemoptysis, occurring either in the context of choriocarcinoma syndrome or as a direct consequence of endobronchial involvement, is often life-threatening. An extremely elevated $\beta$-HCG at presentation (i.e. above 50,000IU/L) is associated with a worse prognosis. However, early recognition, joint management by intensivists, oncologists and urologists, and prompt initiation of cisplatin-based chemotherapy followed by orchiectomy forms the cornerstone of management.

**Acknowledgments**

We acknowledge Dr. Nolan from the Department of Radiology, Kingston General Hospital, Kingston, Ontario, Canada, for providing the chest radiograph and CT scan figures and interpretation.

**References**


