

Primary Pulmonary Choriocarcinoma in a male: a rare case report

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Abstract

Primary Choriocarcinoma (PCC) most commonly occurs in the female genital tract following gestational events such as hydatidiform mole, normal pregnancy, abortion and ectopic pregnancy. The tumor also occurs in males. In males, choriocarcinoma occurs most often in the testes. Less frequently, these tumors have also been reported in other organs such as the urinary bladder, kidney, liver, stomach and colon. Metastasis to the lung is common, but PCC originating in the lung is extremely rare.

The authors report a case of a 26-year-old male non-smoker who presented with respiratory distress and constitutional symptoms along with a large left lung mass and bilateral multiple pulmonary nodules. Clinical and imaging findings of testes and other organs were normal. Level of serum human chorionic gonadotrophin was markedly elevated and it raised the suspicion of primary pulmonary choriocarcinoma of the lung which was confirmed by trans-thoracic biopsy from the mass with immunohistochemistry. This is the third case from India occurring in a male.

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Introduction

Extragenital germ cell tumors (EGCT) account for 2–5% of all germ cell tumors.^[1] Choriocarcinoma in male subjects is one of the non-seminomatous EGCT that occurs in relatively young individuals. Primary (extragenital) choriocarcinoma (PCC) commonly arises in the retroperitoneum, mediastinum and brain.^[2] Metastasis to the lung is common, but PCC originating in the lung is extremely rare. Only seventeen cases have been reported in the world till date.^[3-18] Among these only two are from India.^[8-9]

The prognosis of pulmonary PCC, especially in males is very poor as compared to its testicular counterpart because hematogenous spread is usually common at the time of diagnosis.^[2, 6, 8] Often the entity is misdiagnosed as more common diseases, such as tuberculosis, primary or secondary lung cancer and thus potentially curative chemotherapy or surgery may be delayed.^[6,7,8] So, a high clinical suspicion and a practical approach should be exercised and an urgent biopsy diagnosis is feasible and necessary.

Case Report

A 26-year-old non-smoker male with no significant medical or surgical history presented with complaints of progressive shortness of breath, productive cough, night sweats, diminished appetite and unexplained weight loss which progressive for last 4 weeks. Sputum for AFB was negative. Montoux test was negative. No history of hemoptysis, chest pain, fevers or chills were present. The patient did not respond to routine antibiotics. Chest examination revealed diffuse coarse inspiratory crackles. Genital exam did not reveal any scrotal mass. The remainder of the physical examination was unremarkable.

Laboratory data revealed raised ESR and neutrophilia; normal liver function tests and alpha-fetoprotein (AFP) level. HIV type 1 & 2 were non-reactive.

Chest x-ray showed bilateral pulmonary cannon ball nodules. Computed tomography (CT) demonstrated multiple cannon-ball lesions of variable sizes in both lung fields largest one measuring 8 x 7 x 5 cm in left lung without enlargement of intrapulmonary or mediastinal lymph nodes [Figure 1]. The findings first raised the suspicion of germ cell tumor. Human chorionic gonadotrophin (β -HCG) was markedly elevated (40240 mIU/ml); also lactate dehydrogenase level 870 mg/d. However, FNAC (fine needle aspiration cytology) was suggestive of non-small cell carcinoma possibly squamous cell carcinoma with extensive necrosis.

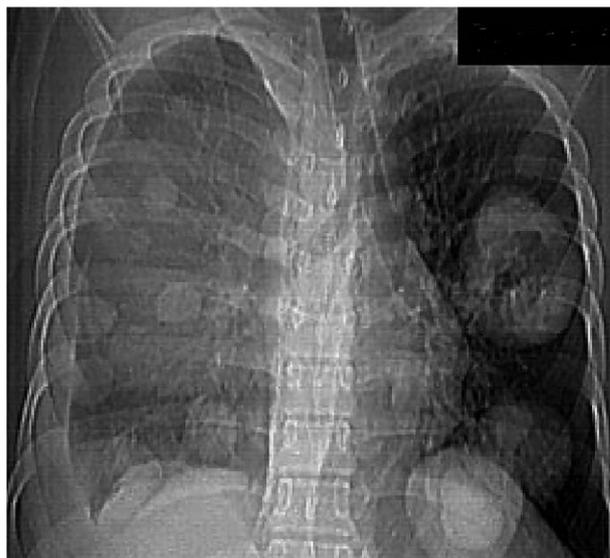


Figure 1: Multiple cannon-ball lesions in both lung fields without enlargement of lymph nodes.

Screening for additional mass especially in the retroperitoneum including CT of the abdomen and brain as well as spine showed no abnormality. CT guided trans-thoracic needle biopsy [Figure 2A] of left lung mass showed large necrotic and hemorrhagic areas with lamellae of large polygonal multinucleated/bizarre cells representing syncytiotrophoblast punctuated by medium to small-sized cytotrophoblasts with clear cytoplasm characteristic of choriocarcinoma [Figure 2B-D]. Immunohistochemistry showed positivity for cytokeratin, β -HCG and negative for PLAP, CD-30 and alpha-feto-protein. There was no retroperitoneal tumor or lymphadenopathy and other organs were also normal.

The patient received four cycles of 20 mg/m² of cisplatin, 100 mg/m² of etoposide and 30 units of bleomycin as per standard protocol. Supportive care was provided as and when necessary. Patient improved markedly following chemotherapy and has been followed up with serial serum β -HCG levels that dropped from 40240 to 720. The patient is now doing well; under therapy and is being monitored.

Discussion

Choriocarcinoma most commonly occurs in the female genital tract following gestational events such as hydatidiform mole, normal pregnancy, abortion and ectopic pregnancy. The tumor also occurs in the absence of pregnancy as well as in men.^[1-6] Primary extragenital choriocarcinoma is rare; usually presenting as a midline lesion in the retroperitoneum, mediastinum, or cranial cavity.^[1, 2, 6,7,8] In men, choriocarcinoma occurs most often in

the testes.^[1] Less frequently, these tumors have also been reported in other organs such as the urinary bladder, kidney, liver, stomach and colon.^[1, 2, 6-8] Accordingly, PCC of the lung is extremely rare. Because the lung is a frequent site of metastasis for choriocarcinoma, a careful search for an occult primary tumor is required though preoperative diagnosis of PCC of the lung is difficult. Not only immunohistochemical studies but also serum levels of HCG have been approved for detection of PCC.^[14, 17, 18]

In present case clinical differential diagnoses like tuberculosis, fungal infections, lymphoma, metastatic carcinoma and small cells or non-small cell carcinomas were effectively ruled out with the help of FNAC and biopsy with immunohistochemistry. FNAC was performed and cytomorphology was suggestive of non-small cell carcinoma possibly squamous cell carcinoma, based on clumps of ghost cells and apoptotic cells.

Necrosis was predominant. However, high β -HCG level and cannon ball deposits in the lungs could not be explained with cytological diagnosis. Further tests and other organs especially retroperitoneum were evaluated and radiologically those were normal. Trans-thoracic needle biopsies from left lower lobe mass confirmed the diagnosis in this case [Figure 1B - D]. Urgently chemotherapy was started and the patient responded well to the standard protocol. Prognosis of PCC of lung according to literature is worse than its testicular counterpart; but our patient is doing relatively well possibly because the tumor was restricted to the lungs only.^[6-8]

Conclusion

PCC of the lung is an extremely rare disease in males that predominantly occurs in young adults. Symptoms and clinical features are variable and non-specific and may be misleading. Screening of the testes and other organs is also necessary as

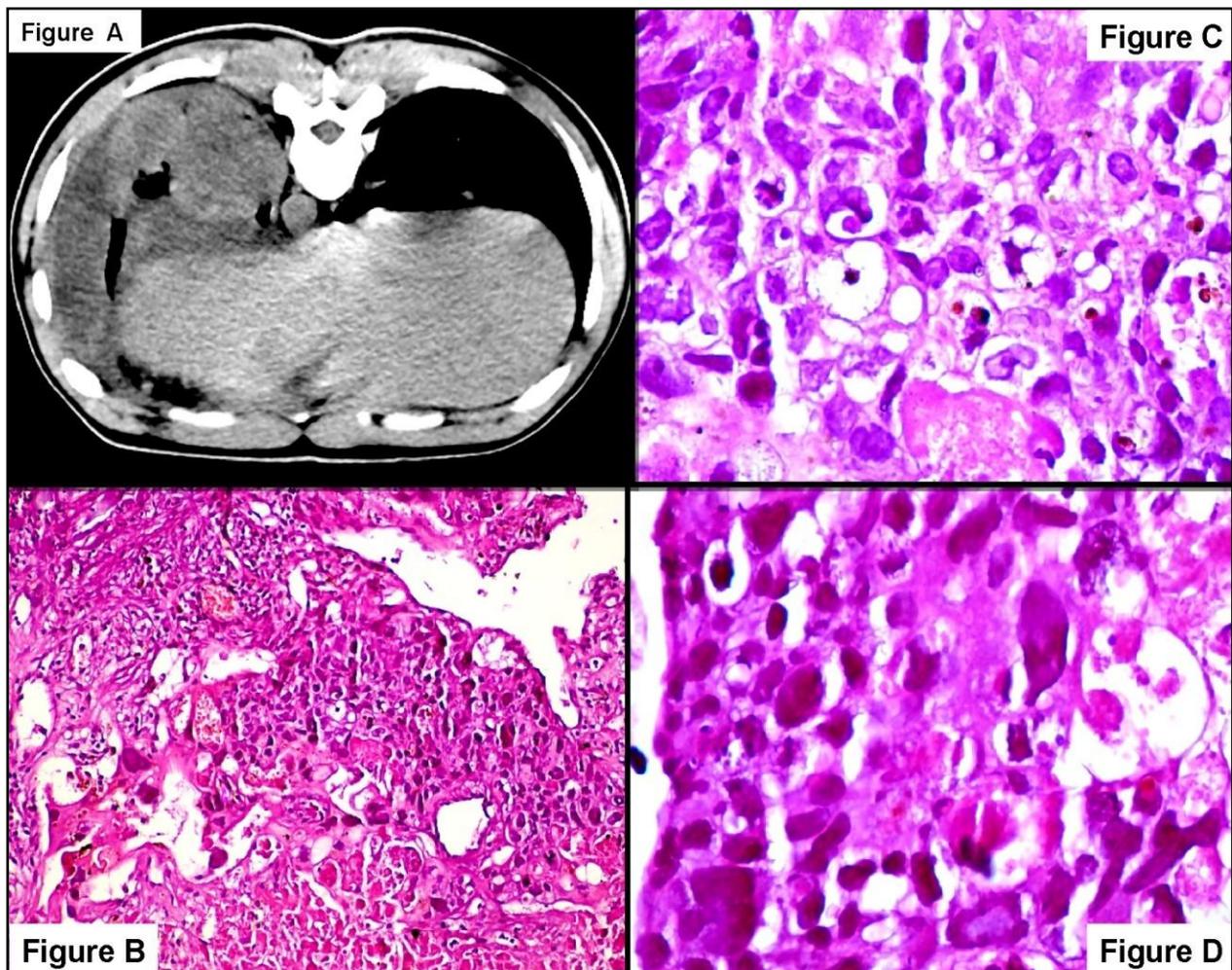


Figure 1A: CT-guided biopsy done from the largest lesion at left lung lower lobe, posterior aspect.
Figure 1B-D: Section showing large necrotic/hemorrhagic areas with lamellae of large polygonal multinucleated/bizarre cells representing syncytiotrophoblast and medium to small sized cytotrophoblasts characteristic of choriocarcinoma. (H & E; 40X & 100X)

secondary pulmonary choriocarcinoma is more common than the PCC. Serum β -HCG level helps in early diagnosis. FNAC and cytological features are those of an undifferentiated highly malignant tumor with Squamous cell carcinoma-like picture. CT-guided trans-thoracic biopsy is an easy, safe and accurate diagnostic procedure. Early institution of cisplatin-based chemotherapy gives comparable results to that of testicular choriocarcinoma especially in the tumor restricted to the lungs only.

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Competing Interests

None declared.

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