

A Case of Pulmonary Artery Pseudoaneurysm Secondary to Epithelioid Choriocarcinoma

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Abstract

Background: Pulmonary Artery pseudoaneurysms (PAP) can cause significant morbidity and mortality in case of a complication and rupture. PAP caused by the malignant tumours is rare.

Case Presentation: We present a case of 35 year old Chinese women who presented with spontaneous left sided chest pain imaging revealed cavitating lesion in the anterobasal segment of her left lower lobe. She underwent thoracotomy and lobectomy. Histology revealed it to be metastatic choriocarcinoma.

Conclusion This case illustrates that metastatic carcinoma could be a possible etiology for PAP. Proper pre-treatment evaluation of PAP and pulmonary artery aneurysms necessitates surgical biopsy to rule out malignancy.

Keywords: thoracics, pulmonary artery, pseudoaneurysm, choriocarcinoma

Background

Pulmonary artery pseudoaneurysms (PAP) are pulmonary artery dilatations not involving all three layers of the vessel wall. They can cause significant morbidity and mortality in the event of rupture. Causes include infection, trauma and malignancy [1]. Metastatic choriocarcinoma as an etiological factor has not been reported. Choriocarcinomas are malignant gestational trophoblastic disease and is more detrimental in the south-east Asian population. We present a case of primary ovarian epithelioid choriocarcinoma with pulmonary metastasis resulted in a PAP.

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Case description

A 35-year old Chinese female presented with one day of pleuritic left sided chest pain. She denied history of trauma or fever. On examination, we found absent breath sounds on her left hemithorax. A moderate left sided effusion was noted on her chest x-ray. She underwent ultrasound-guided aspiration, which revealed markedly bloodstained fluid.

Computerized tomographic angiogram showed a 2.6X2.4 cm size cavitating lesion in the anterobasal segment of her left lower lobe with an adjacent 2.2X2.0 cm early arterial phase homogeneously enhancing lesion. This was supplied by the apical branch of the inferior pulmonary artery and drained to the inferior pulmonary vein (Figure 1). This is representative of a PAP. The pleural effusion was hyperdense which confirmed its hemorrhagic nature. She was offered the options of interventional embolization versus a thoracotomy and

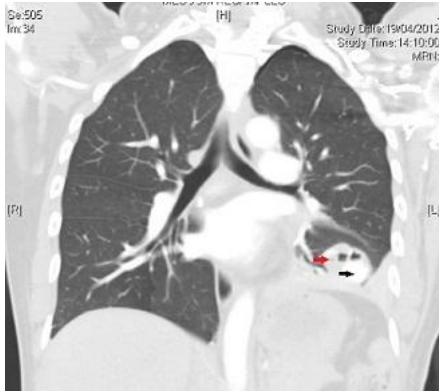


Figure 1: Computerized tomographic angiogram showing the pulmonary artery pseudoaneurysm, supplied by the apical branch of the inferior pulmonary artery and draining into the inferior pulmonary vein. Red arrow indicates tumor, black arrow indicates pseudoaneurysm.

lower lobectomy and opted for the latter. Intraoperatively, we found a 3.0x2x1 cm cystic lesion associated with a 2.2x2.1 cm aneurysm of left lobar pulmonary artery (Figure 2).

The lesion had central necrosis. Viable tissues consisting of irregular islands of polygonal to spindle cells with enlarged, pleomorphic, hyperchromatic nuclei, prominent nucleoli with dense eosinophilic cytoplasm and intracellular bridges were present. Keratinization was present and frequent mitotic activity seen. P63 was found to be positive while CK5/6, CD 31, CD34, and TTF-1 were negative. Initial histological diagnosis of a moderately differentiated keratinizing squamous cell carcinoma was made.

Eight months later, she developed a 4 cm erythematous patch on her occiput and underwent excision. Biopsy of this lesion revealed central necrosis similar to the lung lesion. It had a mixture of large cells with vesicular nuclei, distinct nucleoli with fairly well defined cell margins and syncytial type cells, some multinucleated with hyperchromatic nuclei. There was marked

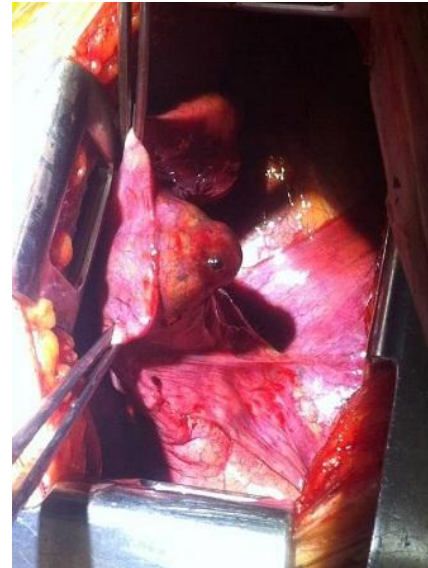


Figure 2: Intraoperative picture of the pulmonary artery pseudoaneurysm

nuclear atypia and high mitotic activity. Cells stained positive for beta human chorionic gonadotropin (β HCG), Human placental lactogen (HPL), AE 1/3, CK 7 and EGFR. Focal positivity for placental alkaline phosphatase (PLAP), EMA inhibin and CD 10 was noted.

Following these findings, the lung resection histology was reviewed and its histological features were similar to the occipital lesion, both being immunopositive for β hCG and hPL. As AS 1/3 and CK7 were also positive, this was determined to be a rare variant of metastatic choriocarcinoma with epithelioid features.

PET CT showed a Flurodeoxyglucose (FDG) avid lesion in her ovary in addition to lung and scalp suggesting that the primary was of ovarian origin. In view of her advanced metastatic disease, she was started on intensive combination chemotherapy.

Discussion

PAPs are a contained rupture of the pulmonary artery. This is in contrast to

aneurysms where all layers of the vessel wall are affected. Etiological factors identified include trauma, malignancy, infection and iatrogenic intervention [1- 3]. Malignant causes include both primary lung cancers and metastasis. However metastatic choriocarcinoma has not been previously documented as an etiological factor for PAP. Various pulmonary involvement of choriocarcinoma are described in literature [4-7]. Due to the rarity of the disease and variable presentation, diagnosis is difficult and retrospective diagnosis of the primary malignancy after detection of pulmonary lesion is also frequently documented [6].

Majority of pulmonary artery aneurysms are treated by watchful waiting, embolization, endovascular stent graft or surgical excision in a form of wedge or lobectomy or surgical vessel ligation. However none of the non-surgical methods provide a histological specimen. As carcinoma is a well-known cause of pulmonary artery aneurysm, evaluation of PAPs for malignancy is important prior to offering catheter-based interventions. At minimum, a PET-CT is necessary to delineate the possible primary sites and other metastatic sites.

Conclusions

This case illustrates that metastatic choriocarcinoma is a possible etiology for PAP. Proper pre-treatment evaluation of PAP and pulmonary artery aneurysms necessitates surgical biopsy or PET-CT to rule out malignancy.

Authors Contributions

HW: Prepared the draft manuscript, literature search.

AAF: preparation of manuscript.

KR: Preparation and editing of manuscript.

AA: Preparation of manuscript.

DBAA: Concept design and editing of the final manuscript for publication.

Ethical Consideration

The written informed consent was taken from the patient for publication of this case report.

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Conflict of interest statement

The authors declare that there are no conflicts of interests. This case was presented at ASCTVS 2014 annual conference.

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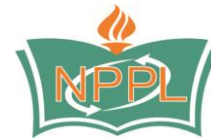
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