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CASE REPORT

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Diffuse alveolar hemorrhage (DAH): a rare presentation of metastatic angiosarcoma

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Abstract

Background Angiosarcoma is an uncommon and highly aggressive malignant tumor. Angiosarcoma presenting as diffuse alveolar hemorrhage (DAH) is rare.

Case presentation A young female who presented with history of dyspnea was found to have features of DAH on radiological evaluation. Angiosarcoma was confirmed from the histopathological examination of the underlying lung nodule. Management with the palliative chemotherapy showed clinical improvement, and resolution of changes of DAH on imaging.

Conclusion Angiosarcomas are not usually listed in the causes of DAH. It must be considered in the differentials of DAH after ruling out the common causes.

Background

Diffuse alveolar haemorrhage (DAH) is a clinical syndrome due to the accumulation of red blood cells within the alveoli, originating from the alveolar capillaries. This condition is characterized by a cluster of symptoms, including hemoptysis, hypoxia, drop in hemoglobin level, and diffuse pulmonary infiltrates on radiographic images [1]. It encompasses various etiologies, ranging from immune-related factors to nonimmune pathologies. Malignancies are not generally considered in the differential diagnosis of DAH, and it is rarely the presenting feature of angiosarcomas.

Case presentation

A 38-year-old female, presented to our institute with history of sudden onset dyspnea associated with hemoptysis. She was initially evaluated elsewhere and referred to our institute for further evaluation. CT chest showed multiple nodules of varying sizes in the bilateral lungs with patchy areas of consolidation and confluent areas of ground glass opacity involving the bilateral lungs possibly DAH (Fig. 1A). Bronchoscopy and lavage confirmed the DAH, and ruled out other infective etiologies. CT guided biopsy from the lung nodule showed plump to spindle cells arranged in sheets with moderate pink cytoplasm with significant mitosis. On IHC tumor cells were positive for CD31, CD34, ERG-1, and FLI-1; while negative for CK, SMA, P40, TTF1, and MDM2. Overall features were suggestive of angiosarcoma. A PET CT scan showed FDG avid right atrial mass (Fig. 1B). After initial supportive care and stabilization, she was started on palliative chemotherapy with weekly paclitaxel and propranolol. Patient had significant clinical improvement on therapy. A re-evaluation CT chest performed after 2 months showed significant decrease in size of lung nodule with resolution of lung infiltrates (Fig. 1C). She received 18 cycles of weekly Paclitaxel, after which she had disease progression along with recurrence of DAH. She was subsequently started on doxorubicin chemotherapy, and had clinical/radiological response.

Angiosarcoma is an uncommon and highly aggressive tumor that develops from endothelial cells, amounts to 1–2% of all soft-tissue sarcomas. These tumors are characterized by their aggressive clinical course, and the

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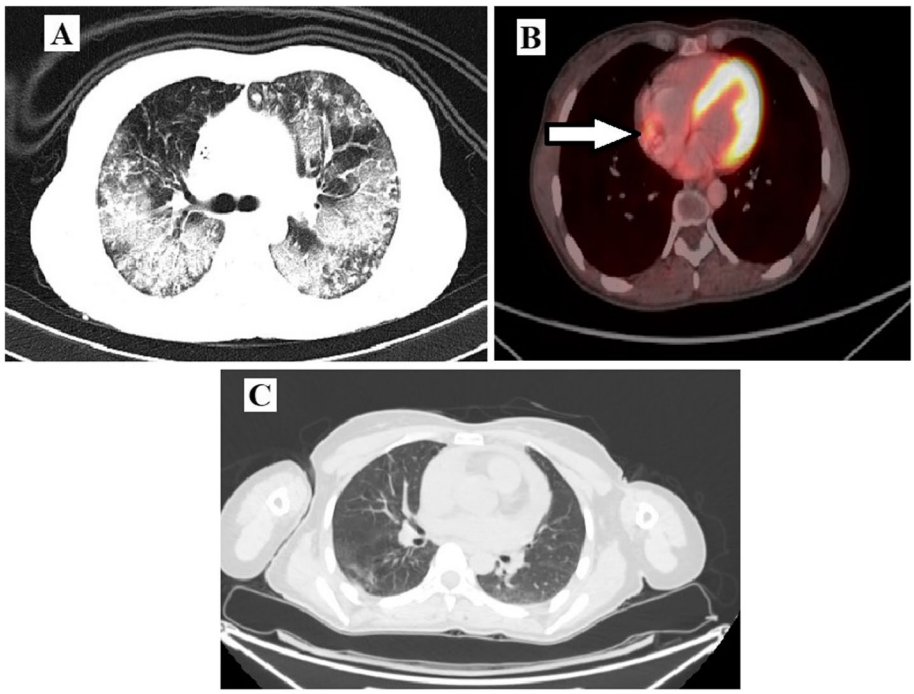

Fig. 1 Initial CT and PET images (**A** and **B**) showing confluent areas of ground glass opacity involving the bilateral lungs and FDG avid right atrial mass. **C** Post-treatment scan showing resolution of initial CT changes

clinical manifestations can vary depending on the anatomic location. DAH is usually caused by connective tissue disorders resulting in capillaritis (immune related). DAH resulting from oncological cause is rare and literature review is limited few case reports [2, 3] These include few cases of angiosarcoma, choriocarcinoma, and renal cell carcinoma. Other differentials in an oncology setting are infections leading to ARDS, coagulopathies. Clinical suspicion of DAH is to be raised based on clinico-radiological suspicion. Patients usually present with dyspnea/hypoxia and hemoptysis. Laboratory investigations show considerable hemoglobin drop. Radiological images show ground glass/consolidative opacities which are usually diffuse and bilateral, and more often central than peripheral [4]. The diagnosis of DAH is confirmed by demonstrating an increasing bloody return on successive aliquots of instilled saline during bronchoalveolar lavage, and hemosiderin laden macrophages in cytological examination of BAL fluid.

Conclusions

Malignancies are not generally considered in the differential diagnosis of DAH. Angiosarcomas can rarely present as DAH. It must be considered in the differentials after ruling out the common causes while evaluating a case of DAH.

Abbreviations

DAH	Diffuse alveolar haemorrhage
PET CT	Positron emission tomography–computed tomography
FDG	Fluoro deoxy glucose
BAL	Bronchoalveolar lavage

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