



Case report

Pulmonary aspergilloma with subcutaneous fistula resulting in massive hemoptysis & subcutaneous emphysema

Anthony Bekasiak*, Aya Shnawa, John Tedrow

St. Elizabeth's Medical Center, Tufts University School of Medicine, Division of Pulmonary, Critical Care & Sleep Medicine, USA

ABSTRACT

Aspergillomas growing in pre-existing lung cavities can lead to presentations of hemoptysis. We present a case of a 73-year-old male with non-small cell lung carcinoma (NSCLC) and known 4 cm left cavitory lesion in the left upper lobe presenting with cough and hemoptysis, leading to the diagnosis of a pulmonary aspergilloma complicated by chest wall subcutaneous emphysema due to cavitory-subcutaneous fistula in the setting of excessive cough. This case sheds light on subcutaneous emphysema as a potential rare complication of aspergillomas in patients without prior trauma.

1. Introduction

Aspergilloma is the most common and most well recognized form of pulmonary involvement due to *Aspergillus* with usual development in pre-existing cavities in the lung. By staying within the borders of a lung cavity, aspergillomas are characterized by their non-invasiveness, as evident in up to 11% of patients who suffered previously from tuberculosis resulting in the formation of cavities [1]. After tuberculosis being the most common associated condition, other cavitory lung diseases can also be complicated by aspergilloma including sarcoidosis, bronchiectasis, bronchial cysts and bulla, ankylosing spondylitis, pulmonary infections and neoplasms [2–4]. Patients with an aspergilloma can be asymptomatic, or may experience mild hemoptysis and even life-threatening massive hemoptysis. This is due to the local hyphal invasion of blood vessels lining the cavity or by mechanical friction of the aspergilloma within the cavitory wall blood vessels [5]. In the presence of massive collateral blood vessels, bronchial artery embolization should be considered as a temporary measure in patients with life-threatening hemoptysis [6].

Pathological causes of subcutaneous emphysema include anaerobic, clostridial, mixed, gas-forming facultative aerobic infections. Other etiologies of subcutaneous emphysema include trauma, iatrogenic and incidental causes, such as barotrauma or labor [7]. Subcutaneous emphysema in the absence of the above conditions appears to be a rare phenomenon. Here, we present a case of an aspergilloma within a previous pulmonary cavity causing a cavitory-subcutaneous fistula without evidence of invasive aspergillosis in the setting of excessive cough resulting in massive hemoptysis and subcutaneous emphysema.

1.1. Presentation of case

A 73-year-old male with severe chronic obstructive pulmonary disease (COPD) with 50 pack year smoking history, coronary artery disease, NSCLC in the left upper lobe treated with chemoradiation and known 4 cm left upper lobe cavitory lesion arrived to our facility with cough and hemoptysis. Vitals were pertinent for tachypnea and hypoxia at 75% room air requiring BiPAP. Lungs were notable for rales and rhonchi bilaterally. He was stabilized and treated for presumed pneumonia and acute exacerbation of his COPD. Of note, he had been evaluated one-week prior at our facility with a similar presentation that resolved with antibiotic administration. His hemoptysis at that time was attributed to his known lung malignancy and dual anti-platelet agents of aspirin and clopidogrel, which were discontinued on discharge. Given his coronary artery disease, and slight drop in hemoglobin, he received two total units of packed red blood cells with appropriate response in his hemoglobin. Chest CTA compared to prior surveillance imaging showed the known cavitory lesion had increased to 4 cm.

At present admission, chest CTA ruled out a pulmonary embolus, but the left upper lobe cavitory lesion increased to 7 cm with increase in soft tissue and decrease in amount of air. CBC showed WBC $21.1 \times 10^3/\mu\text{L}$, hemoglobin 7.0 mg/dL, and platelets of 304 k/uL. Beta-1,3-D-Glucan levels were $< 31 \text{ pg/mL}$, and galactomannan EIA test index < 0.5 . Blood cultures were negative. Sputum cultures were obtained with pending results. The patient continued to experience excessive cough and hemoptysis, therefore with his history of coronary artery disease, he again received blood transfusion with appropriate response in hemoglobin. Given the evolution of the left cavitory lesion, with differential of pus versus blood, he was sent for CT-guided aspiration. In the

* Corresponding author.

E-mail address: anthony.bekasiak@steward.org (A. Bekasiak).

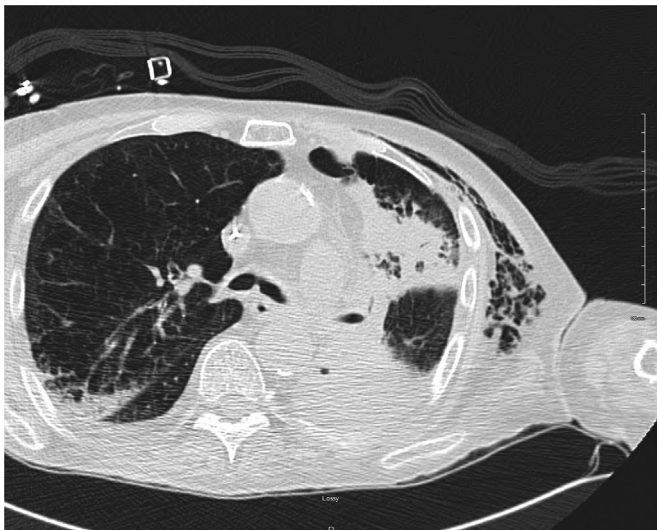


Fig. 1. CT chest for cyst aspiration noting the amount of hyperdense material contained within the left apical cavity significantly decreased, with an air-fluid level now present. In addition, a new finding of moderate subcutaneous emphysema in the left chest wall and axilla are seen. No sub-pleural bleb/bulla was noted. The amount of consolidation in the lingual segment of the left upper lobe had decreased in comparison to imaging two days prior.

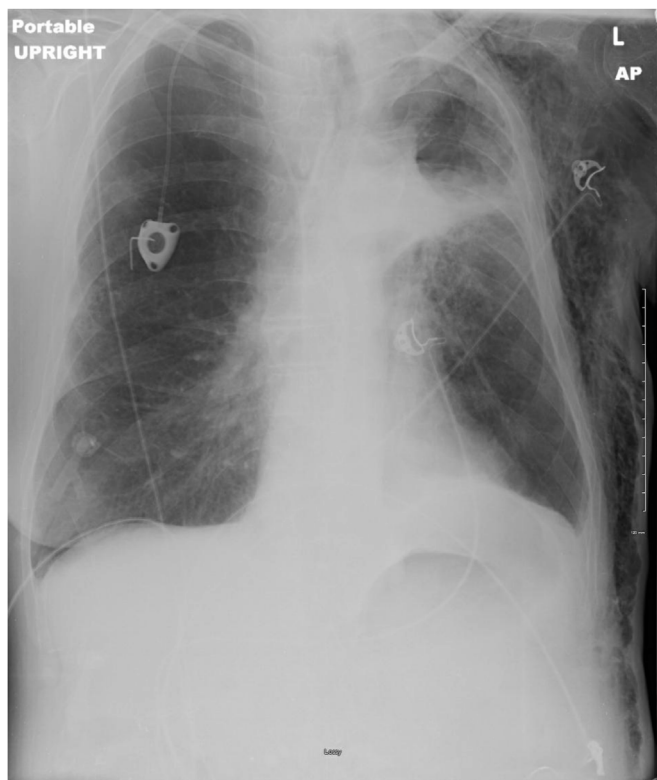


Fig. 2. Portable surveillance chest x-ray demonstrating a stable appearing left upper lung field cavity lesion and extensive left lateral chest wall subcutaneous emphysema.

preprocedural scan, an interval of two days from prior imaging, the amount of hyperdense material contained within the cavity had significantly decreased with an air-fluid level present. Furthermore, a moderate subcutaneous emphysema in the left chest wall and axilla were noted, raising the question of a pleuro/broncho-cutaneous fistula between the air-filled cavity and tissues. No sub-pleural bleb/bulla was noted. Lastly, the amount of consolidation in the lingual segment of the

left upper lobe had decreased in comparison to imaging two days prior (Fig. 1). Surveillance chest x-ray imaging afterwards showed no pneumothorax, and no change in the cavity lesion size, with no significant changes in the subcutaneous emphysema along the left lateral chest wall (Fig. 2) Aspirate cultures returned with heavy growth of *Aspergillus Fumigatus*, along with its growth in the fungal cultures. He was started on voriconazole therapy and transferred to an outside facility for interventional radiology embolization due to continued massive hemoptysis.

2. Discussion

We present a case of an aspergilloma within a previous pulmonary cavity causing a cavity-subcutaneous fistula without evidence of invasive aspergillosis in the setting of excessive cough resulting in massive hemoptysis and subcutaneous emphysema. Subcutaneous emphysema secondary to tuberculosis may develop due to associated pneumothorax, pneumomediastinum, or following chest tube insertion [8]. For it to develop without pneumothorax or pneumomediastinum appears to be a rare complication, with literature review revealing two other accounts as reported by Dixit and Pandey et al. [9,10] Both reports were of adult males with heavy smoking histories, aged 42 and 60 respectively, with presentations of sudden onset shortness of breath after bouts of excessive coughing. Initial chest x-ray imaging in both cases noted diffuse subcutaneous emphysema with right upper lobe cavity lesions, and ensuing CT chests noting demonstrable fistulas. In the case of Dixit et al., a cavernous-pleuro-soft tissue fistula was noted to be communicating to the subcutaneous tissue from the right upper lobe cavitation, and in Pandey et al., a broncho-cavity-subcutaneous fistula was seen. As opposed to an aspergilloma, the culprits in both cases were secondary to pulmonary tuberculosis.

The similarities between all three cases are noteworthy. Fistula communications between subcutaneous tissues arising from pulmonary cavity lesions is noted in all three cases. Moreover, each presentation appears to be in the setting of excessive cough. With an increase in intrapulmonary pressures from excessive coughing, a rupture at a weakened point – for example, at the cavity-subcutaneous fistula in our patient – would allow for air to enter the tissue, resulting in subcutaneous emphysema.

To the best of our knowledge, a case of a non-invasive aspergilloma within a pulmonary cavitation complicated by massive hemoptysis and subcutaneous emphysema without pneumothorax or pneumomediastinum has not been reported. Aside from tuberculosis, staphylococcal pneumonia, measles, pneumocystis carinii, influenza pneumonia and pertussis are other infections to have caused subcutaneous emphysema, especially amongst children [11]. The treatment of subcutaneous emphysema in many cases are self-limiting and the underlying disease or complication should be addressed and treated. If in hemodynamic instability or respiratory distress, tracheostomy and skin incisions are required to treat subcutaneous emphysema. With massive hemoptysis, a systemic approach is key to timely control and reversal of potentially fatal complications.

Conflicts of interest

None.

Acknowledgements

We would like to thank Dr. John Tedrow in his guidance in this case, alongside Dr. David Lee of Department of Radiology and Dr. Jorge Fleisher of the Infectious Disease Department.

References

- [1] Tuberculosis Association, *Aspergilloma and residual tuberculous cavities*. The

- results of a resurvey, *Tubercle London* 51 (3) (1970) 227–245.
- [2] S. Kawamura, S. Maesaki, K. Tomono, T. Tashiro, S. Kohno, Clinical evaluation of 61 patients with pulmonary aspergilloma, *Intern. Med.* 39 (2000) 209–212.
- [3] C.A. Kauffman, Quandary about treatment of aspergillomas persists, *Lancet* 347 (1996) 1640.
- [4] G. Zizzo, A. Castriota-Scanderbeg, N. Zarrelli, G. Nardella, J. Daly, M. Cammisa, Pulmonary aspergillosis complicating ankylosing spondylitis, *Radiol. Med.* 91 (1996) 817–818.
- [5] Adilia Warris, The biology of pulmonary *Aspergillus* infections, *J. Infect.* 69 (S1) (2014) S36–S41.
- [6] R. Uflacker, A. Kaemmerer, P.D. Picon, et al., Bronchial artery embolization in the management of hemoptysis: technical aspects and long-term results, *Radiology* 157 (1985) 637–644.
- [7] N. Patel, S.K. Lazow, J. Berger, Cervicofacial subcutaneous emphysema: case report and review of literature, *J. Oral Maxillofac. Surg.* 68 (8) (2010) 1976–1982.
- [8] Spontaneous cervical and mediastinal emphysema, G.S. Parker, D.A. Mosborg, R.W. Foley, *Stiernberg CM Laryngoscope* 100 (9) (1990 Sep) 938–940.
- [9] R. Dixit, J. George, Subcutaneous emphysema in cavitary pulmonary tuberculosis without pneumothorax or pneumomediastinum, *Lung India : Off. Organ Ind. Chest Soc.* 29 (1) (2012) 70–72, <https://doi.org/10.4103/0970-2113.92369>.
- [10] Deveshwar Pandey, Pramod Jaret, Rajesh Sharma, Amit Sharma, Surinder Thakur, Subcutaneous emphysema secondary to pulmonary cavity in absence of pneumothorax or pneumomediastinum, *Respir. Med.* 101 (2) (2007) 363–365.
- [11] M. Das, U.C.M. Natchu, R. Lodha, S.K., Kabra Pneumomediastinum and subcutaneous emphysema in acute miliary tuberculosis, *Indian J. Pediatr.* 71 (2004) 553–554.