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Subcutaneous Emphysema and**Pneumomediastinum: An Unusual Presentation of Lung Cancer**

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Abstract

Lung cancer in women has significantly increased during the last forty years. Lung adenocarcinoma is diagnosed at an average age of 71 years, and it is extremely uncommon for this type of cancer to be diagnosed before the age of 20. Subcutaneous emphysema with underlying pneumomediastinum are exceedingly unusual presentations of lung neoplasms and till now only eleven cases have been reported worldwide. Here we mention a case of adenocarcinoma in a young female presenting with subcutaneous emphysema and pneumomediastinum. Although the prognosis of lung cancer with such atypical features are usually poor, our patient showed dramatic improvement owing to early diagnosis and management of the tumor.

Introduction

The leading cause of cancer-related deaths in women is lung cancer. Lung cancer kills more women annually than breast, ovarian, or uterine cancer combined [1]. 16% of women who develop the most common types of lung cancer have never smoked, despite the fact that smoking is the main cause of the disease [2]. There are two types of lung cancer, namely non-small cell lung cancer (NSCLC) and small cell lung cancer (SCLC). As cigarette smoking is the primary cause of SCLC, it is more frequent among men. [3]. On the contrary, women may present NSCLC in either of the two forms: adenocarcinoma or squamous cell carcinoma [2].

Female patients with lung adenocarcinoma frequently with symptoms such as dyspnoea, cough, hemoptysis, chest pain, fatigue, back or shoulder pain [4]. Spontaneous pneumomediastinum leading to subcutaneous emphysema is one of the rarest presentations of lung cancer. Till now only a few cases of lung neoplasms presenting with mediastinal emphysema have been reported [5]. The collection of air in the mediastinum, known as pneumomediastinum, can be caused by a number of triggering causes, including persistent coughing, vomiting, or even substance abuse [6]. It is extremely rare for lung cancer patients to experience pneumomediastinum. The air buildup in the mediastinum is either due to a fistula in the digestive tract or a leak from the tracheobronchial tree, usually as a result of destruction of the lung parenchyma caused by the neoplasm itself [7]. Subcutaneous emphysema may result from an underlying pneumomediastinum, as excessive accumulation of air can migrate into the subcutaneous tissues through the damaged tissue [8].

Here, we present a rare case of lung adenocarcinoma presented with

subcutaneous emphysema and pneumomediastinum in a 13 year old female patient. As far as we are aware, this is the first instance of lung cancer in Bangladesh to be detected with such rare clinical presentation.

Case Report

A 13 years old female presented with dry cough for 3 months, haemoptysis for 2 months, and shortness of breath for 5 days along with history of face and neck swelling for the same duration. There was no history of chest trauma or any surgical procedure. She did not have any history of food or drug allergy.

She was seated for improved ventilation owing to respiratory distress and required oxygen support. Crackles on her right and left haemithorax, extending to the neck, were found on chest examination. On the contrary, the breath sounds were audible from both sides. Complete blood count showed haemoglobin 9.6 g/dl, erythrocytic sedimentation rate (ESR) 60 mm in 1st hour and total count of white blood cells $11.5 \times 10^9/L$ (Neutrophil- 67%, Lymphocyte- 28%). Sputum was negative for acid-fast bacilli.

Chest radiography displayed subcutaneous emphysema as indicated by the red arrow (Figure-1). Computed tomography (CT) scan of the chest showed pneumomediastinum, subcutaneous emphysema along with a soft tissue mass (indicated by the yellow arrow in Figure-2). Bronchoscopic examination was conducted which further revealed a vascular fleshy mass in the left principal bronchus, 2 cm from the carina. The mass was inflamed and multiple haemorrhagic spots were observed. Endobronchial ablation of the tumour using a Nd:Yag laser was performed. Adenocarcinoma was diagnosed through histopathological analysis of the biopsy material taken during the operation. The patient was referred to an oncologist for additional care once the emphysema resolved.

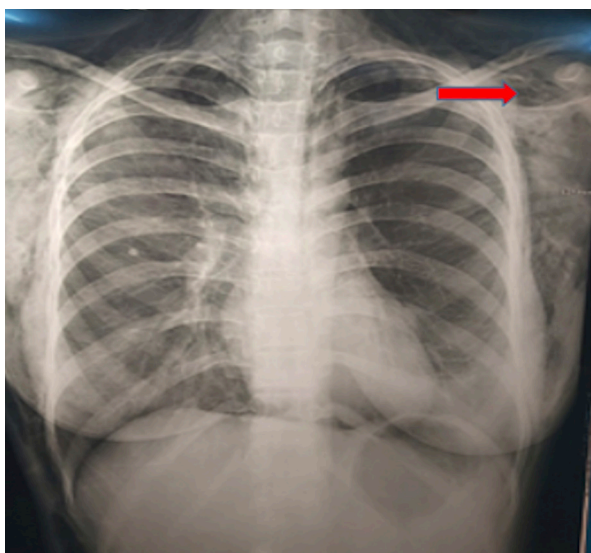


Figure 1: Chest Radiography showing subcutaneous emphysema as indicated by the red arrow



Figure 2: Chest CT reveals subcutaneous emphysema and mass in the trachea (as indicated by the yellow arrow).

Discussion

Non-small cell lung cancer (NSCLC) accounts for 85% of all occurrences of lung cancer, making it the leading cause of cancer-related death worldwide [1]. Adenocarcinoma is the most prevalent type of lung cancer in patients; in the US, it accounts for about 30% of cases in men and 37% in women [9]. Spontaneous pneumomediastinum with subcutaneous emphysema are exceedingly rare presentations of lung adenocarcinoma. Till, 2023 around eleven such cases have been reported, of which seven have been described in a brief review by Barquero-Romero and Redondo-Moralo [5]. The four other cases with such atypical presentations and outcome have been tabulated [8, 10-12]. Interestingly, all eleven patients with similar features are aged above 45 years, whereas the patient described is aged only 13. Hence, lung neoplasms must be ruled out in younger patients presenting with pneumomediastinum, pneumothorax or subcutaneous emphysema.

The underlying mechanism of subcutaneous emphysema in lung neoplasms involves accumulation of air in the fascial planes either due to an abnormal communication between the plane and a structure normally in contact with air or due to destruction of parenchymal tissue or the ribs by the neoplasm itself [7,13]. In this case, bronchoscopic examination revealed an inflamed fleshy mass in left principal bronchus with multiple hemorrhagic spots. On ablation of the mass, the patient's condition improved, suggesting that the emphysema was probably caused due to pressure necrosis of the lung parenchymal tissue. No fistula was detected between the subcutaneous tissue and an air-filled organ.

Pneumomediastinum is usually managed with conservative treatment that includes the use of analgesics, oxygen supplementation and a course of antibiotic in case of mediastinitis [8]. Symptomatic subcutaneous emphysema may require the insertion of a large bore drain [14]. However, oxygen supplementation along with removal

Table 1: Summary of presenting complaints, investigations and outcome of cases of lung adenocarcinoma presented with subcutaneous emphysema and pneumomediastinum

Age and gender	Presenting complaints	Investigation findings	Spontaneous Pneumo-mediastinum	Subcutaneous emphysema	Treatment	Outcome	Reference
45 y/o; male	Development of bronchopleural fistula following left upper lobe wedge resection for lung adenocarcinoma	Chest CT scan revealed pneumomediastinum causing tracheal compression	Yes	Yes	Conservative management with chest tube suction	Recovered	[10]
47 y/o; female	Dyspnoea; choking sensation; facial and neck swelling	Bronchoscopy revealed endotracheal obstructing mass	Yes	Yes	Endobronchial ablation of the tumour using a Nd:Yag laser	Recovered	[11]
69 y/o; male	Tachypnoea; cervicofacial swelling	Chest CT scan revealed destruction of ribs with subcutaneous emphysema	Yes	Yes	Insertion of drainage tubes	Recovered	[12]
79 y/o; female	Diagnosed case of stage IVB cancer of the left lung with metastasis	Thoraco-abdominal CT scan revealed subcutaneous emphysema from the cervical to the pelvic region, extensive pneumomediastinum and pneumopericardium	Yes	Yes	Conservative management	Deceased	[13]

of the mass dramatically improved the condition of our patient and a drainage tube was not required.

The case studies suggest that the overall prognosis of lung carcinoma with such atypical presentations is extremely poor with a case fatality rate of 50% within 30 days. Patients with advanced stage of lung carcinoma usually presents with pneumomediastinum, pneumothorax, pneumoperitoneum or subcutaneous emphysema. The rapid recovery of our patient may attribute to the presentation of rare clinical features at an early stage of the neoplasm as well as prompt diagnosis and management. Consequently, airway malignancies should be suspected in patients of any age exhibiting subcutaneous emphysema and pneumomediastinum, and managed accordingly.

Conclusion

Adenocarcinoma of lung is common in younger female patients. When a young patient presents with unusual clinical characteristics, bronchial adenocarcinoma should be taken into account in the differential diagnosis. Delay in diagnosis can result in potential complications like metastasis and death. High level of suspicion along with targeted clinical approach can lead to an early diagnosis and positive outcome.

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Rahman Mia and Dr. Md. Monirul Islam contributed to case selection and critical revisions of the manuscript.

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Consent: The authors certify that they have obtained all appropriate patient consent forms. In the form the patient's guardian has given his consent for the patient's clinical information to be reported in the journal. The patient and her guardian understand that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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