



# Spontaneous Pneumomediastinum in a Patient with Allergic Bronchopulmonary Mycosis

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## ABSTRACT

Spontaneous pneumomediastinum (SPM) is the manifestation of air in the mediastinum in the non-attendance of trauma and it is uncommon. We showed spontaneous pneumomediastinum, a rare complication after an asthma attack. ABPM is a rare disease, often associated with severe asthma. Fungal sensitization is an issue in severe asthma and this type of sensitization is important in terms of drawing attention to different complications in a case. Although the patient's mold and yeast mixture (Mx2) allergen-specific immunoglobulin E-ImmunoCAP system (Phadia 100, Phadia, Uppsala, Sweden) was positive, no susceptibility to aspergillus was detected in specific IgE and skin tests. Total IgE was 2560 kU/L. Our asthmatic patient with candida albicans and penicillium sensitivity on skin test was diagnosed with allergic bronchopulmonary mycosis (ABPM). This case highlights the importance of raising awareness that there may be an increased risk of SPM in Asthmatics with ABPM.

**Keywords:** Spontaneous pneumomediastinum, mold sensitivity, severe asthma, allergic bronchopulmonary mycosis ABPM

## INTRODUCTION

Asthma is one of the most widespread chronic respiratory diseases. Patients with allergic bronchopulmonary mycosis (ABPM) present with clinical features of ABPA, but skin and serological tests against *Aspergillus fumigatus* are negative.

Negative prick and intradermal tests along with low level of serum specific IgE tests against aspergillus nearly exclude ABPA, yet high Total IgE levels and severe asthma may indicate sensitization against other molds like penicillium or alternaria etc.

Mold sensitization is significantly related with severe asthma. It has been previously shown that patients with *Alternaria* susceptibility have a 14-fold greater risk of SPM and a greater risk of fatal asthma aggravations. It is put forward that molds, unlike other allergens, act through a specific mechanism called the 'air leaking effect' (1). SPM is the manifestation of air in the non-attendance of trauma

and it is uncommon. Triggers for SPM include respiratory infections and maneuvers (e.g., vomiting, Valsalva, strenuous exercise). Severe asthma attacks are one of these predisposing causes. It is usually a self-limiting entity with spontaneous resolution and conservative follow-up (2-5).

We presented a case of an elderly female patient who presented to the emergency department with a severe asthma attack. When we evaluated the patient, we diagnosed the patient with ABPM. During the follow-up period with conservative treatment for SPM in addition to the asthma attack treatment, spontaneous remission of SPM was achieved.

It is well known that patients with ABPA or ABPM have a more severe degree of disease than patients with other asthmatics (6). Based on this information, care should be taken in terms of SPM, which is a serious complication of attacks in these patients. In a patient describing increasing dyspnea, solely an attack should not be considered as the cause of dyspnea, and the patient should be checked

for SPM radiologically. We present SPM as an important complication of a patient with ABPM, a special group of patients with severe asthma.

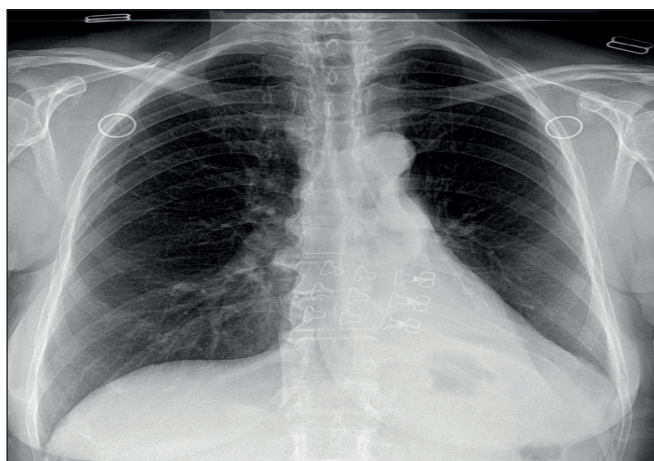
With this case, it is aimed to draw attention to a severe unexpected complication of this group.

### CASE HISTORY

A 63-year-old female patient, known to have asthma for 30 years, applied to the emergency department with the complaint of shortness of breath and cough. The patient had no history of surgery or trauma. On admission, physical examination showed that blood pressure was normal; respiration was 28 per min with room air and oxygen saturation of 86%; body temperature was 36.4°C. The patient had a pulse of 106/min and bilateral rhonchi, with prolonged expiration. Other system examinations were unremarkable.

ECG showed sinus rhythm. Considering that the first chest X-ray was normal, a severe asthma attack was considered according to the physical examination and clinical findings, and asthma attack treatment consisting of salbutamol, glucocorticoid, and oxygen support was applied. 2 g intravenous magnesium sulfate was given to the patient who did not feel relief for the first 2 hours. The patient who did not respond to this treatment was consulted with the chest diseases, thorax CT was requested and SPM was detected in the patient.

As shown in Figure 1, Chest X-ray on admission and as illustrated in Figure 2, Chest computed tomography (CT)



**Figure 1.** Chest X-ray of the case disclosing air within the mediastinum.

were performed for our patient because we suspected a complicated asthma attack.

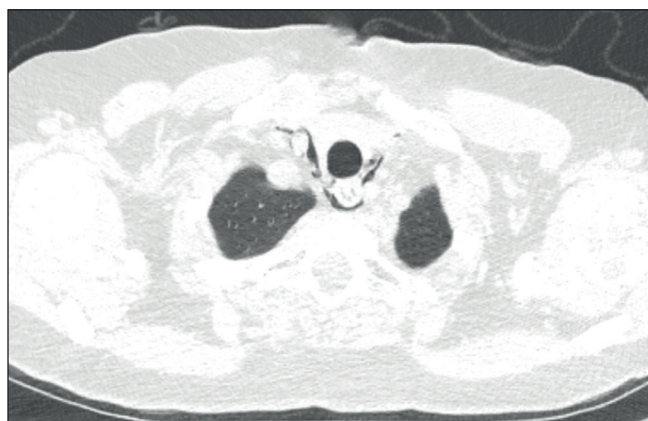
Chest CT analysis displays that there was at the pre-vascular level free air densities in favor of pneumomediastinum in the mediastinum, trachea, and in the traces of both main bronchi, and in the paraesophageal area. There was total atelectatic volume loss in the lower lobe of the left lung.

There was no obvious sign of pneumonic infiltration in the lungs.

Arterial blood gases while receiving 2-3 lt/min oxygen therapy demonstrated mild hypoxemic respiratory alkalosis. (pH, 7.51; pCO<sub>2</sub>, 31.9 mmHg; pO<sub>2</sub>, 70.2 mmHg; base excess 2.5 mmol/L; lactate 4.7mmol/L; and, HCO<sub>3</sub><sup>-</sup>: 25.4mmol/L). The patient's troponin values were normal, and other related blood tests were not noteworthy. The total eosinophil counts was 910 /mm<sup>3</sup>.

The patient, who was diagnosed with severe asthma attack according to the 2020 national asthma diagnosis guideline, was hospitalized with the diagnosis of severe asthma attack and SPM. A thoracic surgeon was consulted for SPM. Conservative treatment was recommended to the patient. Bronchoscopy was performed for left lung lower lobe atelectasis. In bronchoscopy, no pathology was observed in the atelectatic left lower lobe. Bronchoscopy showed that the lower lobe was open and ventilated.

The patient's complaints regressed after oxygen and steroids (5 days), and X-ray was taken again before dis-



**Figure 2.** CT analysis displaying air within the mediastinum, trachea, and in the traces of both main bronchi, and in the paraesophageal area.

charge. As demonstrated in Figure 3, It was observed that the pneumomediastinum was almost completely healed.

The patient was discharged after SPM remission was observed in the control chest X-ray at the first week of her hospitalization.

The patient's asthma was under full control at the outpatient clinic control on the 15th day.

General atopy evaluation, sIgE, and skin prick tests were performed on the patient who had severe asthma attacks twice in the last year. Mold sensitivity was investigated in detailed tests because of the severe asthma attack, allergic rhinitis, high total IgE of 2560 kU/L, and feNO of 35 ppb. In the patient with type 2 inflammation clinic, mold and yeast mixture (Mx2) allergen-specific immunoglobulin E was 4.01 (+++) kUA/L. In detailed tests, mold susceptibility was especially investigated and skin tests were positive for candida (3x3 mm in prick test and 9x9 mm in intradermal 1/1000 test) and penicillium (4x5 mm in intradermal 1/10 test). The skin prick test against *Aspergillus* and *Alternaria* was negative. M3 *Aspergillus fumigatus* was 0.12 (neg) (<0.35 kUA/L), m6 (*Alternaria alternata*) specific IgE was 0.07 kUA/L, and *Aspergillus Mix (mx4)* specific IgE was 0.09 kUA/L.

This case presents a clinically mold-sensitive patient with more severe and frequent exacerbations in the severe asthma group. As a matter of fact, SPM, which is a rare complication, developed in our patient.

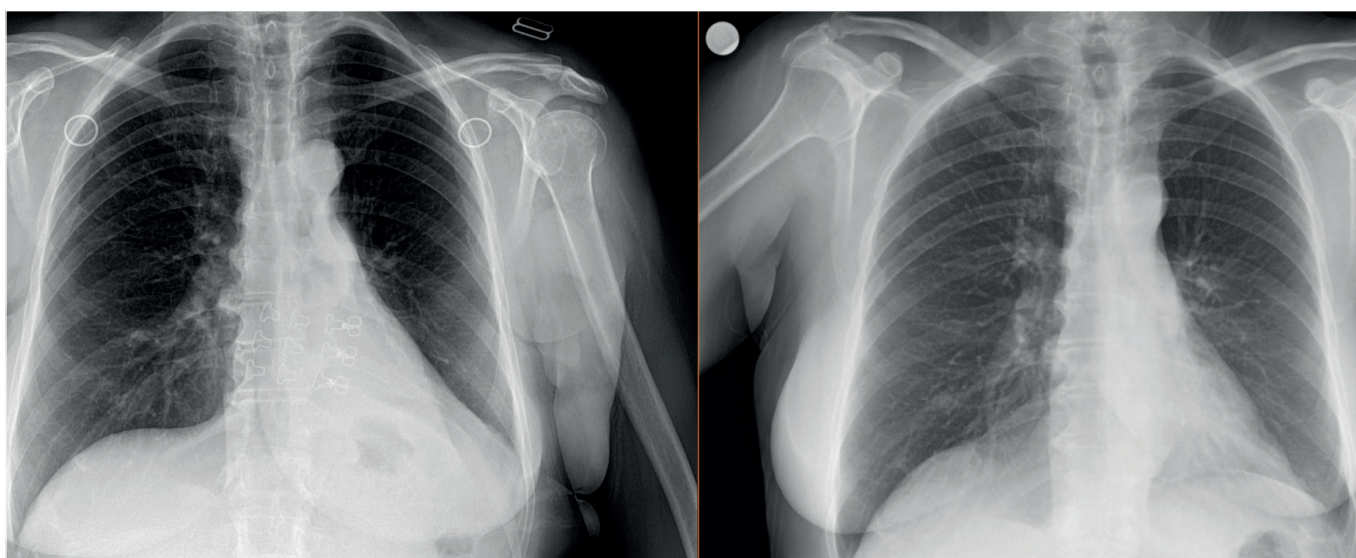
As in our case, it is always necessary to be alert for SPM, which is a clinical entity that can easily be overlooked with a chest X-ray.

## DISCUSSION

SAFS (Severe asthma with fungal sensitization) and ABPA are clinical conditions caused by the immune response due to fungal sensitization in severe asthma. ABPA/AMPM may develop over time in patients with SAFS. Fungal susceptibility to *aspergillus* is most common in severe asthma (7). In the literature, a 14-fold increase in risk has been shown for SPM in *Alternaria* (4). Our case was ABPM susceptible to *candida* and *penicillium*.

This case characterized spontaneous pneumomediastinum, a rare complication after an asthma attack. Although spontaneous pneumomediastinum is very rare (1:7000 to 1:45000), it should not be forgotten, especially in those with chest pain and shortness of breath (8). Although SPM is generally a benign condition in patients with asthma, it may occur secondary to radiotherapy in patients with lung cancer (9).

Chest pain is the most common manifestation in most cases. The other most extensive symptoms are dyspnea, cough, and subcutaneous emphysema, respectively. Hamman's sign is a crackling or rasping sound that is synchronous with the heartbeat. It exists in roughly a fifth of the patients (10). We could not detect Hamman's sign or subcutaneous emphysema on clinical examination.



**Figure 3.** First chest radiograph on the left, chest radiograph at discharge on the right.

There are three principal elements of the therapy of SPM: oxygen therapy, relaxation, and analgesia (11). We treated our case with oxygen and prednol (40 mg/5 days), and in our case things went very well and we were successful.

We showed that our patient had sensitivity to *Candida albicans* and penicillium. We recommend larger studies to be conducted to detect the increased SPM risk with other molds.

## CONCLUSIONS

This case highlights the importance of raising awareness that there may be an increased risk of SPM in Asthmatics with ABPM.

Further observation is required with new studies to determine the frequency of the association between ABPA/ABPM and SPM.

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## Authorship Contributions

Concept: **Kasım Okan**, Design: **Kasım Okan**, **Meryem Demir**, Data collection or processing: **Kasım Okan**, **Meryem Demir**, **Hatice Serpil Akten**, Analysis or Interpretation: **Ozlem Goksel**, Literature search: **Kasım Okan**, **Meryem Demir**, **Hatice Serpil Akten**, Writing: **Kasım Okan**, **Ozlem Goksel**, Approval: **Kasım Okan**, **Ozlem Goksel**.

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