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Clinical Manifestations of Spontaneous Pneumomediastinum

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Background: Spontaneous pneumomediastinum (SPM) is an uncommon disorder with only a few reported clinical studies. The goals of this study were to investigate the clinical manifestations and the natural course of SPM, as well as examine the current available treatment options for SPM. **Methods:** We retrospectively reviewed 91 patients diagnosed with SPM between January 2008 and June 2015. **Results:** The mean age of the patients was 22.7±13.2 years, and 67 (73.6%) were male. Chest pain (58, 37.2%) was the predominant symptom. The most frequent precipitating factor before developing SPM was a cough (15.4%), but the majority of patients (51, 56.0%) had no precipitating factors. Chest X-ray was diagnostic in 44 patients (48.4%), and chest computed tomography (CT) showed mediastinal air in all cases. Esophagography (10, 11.0%), esophagoduodenoscopy (1, 1.1%), and bronchoscopy (5, 5.5%) were performed selectively due to clinical suspicion, but no abnormal findings that implicated organ injury were documented. Twelve patients (13.2%) were discharged after a visit to the emergency room, and the others were admitted and received conservative treatment. The mean length of hospital stay was 3.0±1.6 days. There were no complications related to SPM except for recurrence in 2 patients (2.2%). **Conclusion:** SPM responds well to conservative treatment and follows a benign natural course. Hospitalization and aggressive treatment can be performed in selective cases.

Key words: 1. Spontaneous pneumomediastinum
2. Mediastinal emphysema
3. Outpatients

Introduction

Pneumomediastinum (PM) is defined as the presence of air or other gas in the mediastinum; it is also known as mediastinal emphysema [1]. It can be categorized as spontaneous PM (SPM) or secondary PM [1]. SPM is a rare and benign disorder that generally occurs in young adult males without any precipitating factors or disease [1,2]. The hypothesized pathogenesis of SPM was described as the Macklin effect in 1944 [1-3]. Alveolar rupture might lead to air dissection along the bronchovascular sheaths, leading to pulmonary interstitial emphysema that spreads into the mediastinum [2-4]. Secondary PM has a definite precipitating factor such as trauma, a surgical or medical procedure (iatrogenic PM), or infection by a gas-forming organism [5,6].

Generally, SPM is considered a comparatively benign disease, but few studies have been published covering substantial data pertaining to SPM [2,4,7]. Previous reports on SPM are usually case series of small numbers of patients; therefore, the clinical manifestations of SPM have not been fully elucidated due to the rarity of this condition [7]. The aim of this study was to document the clinical features and course of SPM and identify the optimal diagnostic methods and treatment by analyzing a large number of patients with SPM.

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Table 1. Patients' symptoms

Symptoms	No. of patients (%)
Chest pain	58 (37.2)
Cervical pain	28 (17.9)
Dyspnea	15 (9.6)
Cough	12 (7.7)
Crepitus sensation	10 (6.4)
Dysphagia	9 (5.8)
Febrile sensation	7 (4.5)
Back pain	5 (3.2)
Dysphonia	3 (1.9)
Abdominal pain	3 (1.9)
Pharyngeal enlargement	2 (1.3)
Dizziness	1 (0.6)
Rhinolalia	1 (0.6)
Facial swelling	1 (0.6)
General weakness	1 (0.6)

Table 2. Triggering events

Triggering events	No. of patients (%)
None	51 (56.0)
Cough	14 (15.4)
Diet	13 (14.3)
Sports (Valsalva maneuver)	7 (7.7)
Vomiting	3 (3.3)
Shouting	1 (1.1)
Working	1 (1.1)
Blowing	1 (1.1)

Methods

In total, 162 patients with PM were identified during the study period (January 2008 to June 2015). SPM was defined as the radiologic confirmation of air within the mediastinum without any underlying factor. The exclusion criterion was PM with a definite precipitating factor such as trauma, a surgical or medical procedure (iatrogenic PM), or infection. Finally, we retrospectively reviewed 91 patients who were diagnosed with SPM. All medical records of these patients were reviewed with consideration of etiologic factors (age and sex), symptoms, precipitating factors, trigger events, complications, radiologic findings, offered treatment, length of hospital stay, natural course, and outcome. Categorical variables are expressed as percentages, and continuous variables are expressed as mean±standard deviation. This study was approved by the institutional review board of Ajou University Hospital (MED-MDB-15-268).

Table 3. Predisposing conditions

Condition	No. of patients (%)
Smoking	19 (20.9)
Upper respiratory infection	7 (7.7)
Asthma	3 (3.3)
Chronic obstructive pulmonary disease	1 (1.1)
Drugs	0

Results

Among the 91 patients with SPM, the mean age was 22.7±13.2 years (range, 12–78 years), and 67 (73.6%) patients were male. The most frequently reported symptoms were chest pain (58, 37.2%) and cervical pain (28, 17.9%), followed by dyspnea (15, 9.6%), cough (12, 7.7%), and crepitus sensation (10, 6.4%) (Table 1). If the patient had more than one symptom, all symptoms were described. The mean white blood cell (WBC) count, percent neutrophils, and C-reactive proteins were 9,780±2,930/㎕, 68.1%±10.3%, and 0.5±1.2 mg/dL, respectively. The arterial blood gases were analyzed in 15 patients who complained of dyspnea. The mean partial pressure of oxygen (PaO2) was 93.2±24.1 mmHg.

In 51 patients (56.0%), there was no apparent trigger factor to generate SPM. However, in the remaining 40 patients (44.0%), there were obvious triggering events. SPM developed most frequently during cough (14, 15.4%). Other factors included diet (13, 14.3%) and physical activity, which was related to the Valsalva maneuver (7, 7.7%) (Table 2).

A medical history predisposing the occurrence of SPM included smoking in 19 patients (20.9%), recent upper respiratory infection in 7 patients (7.7%), asthma in 3 patients (3.3%), and chronic obstructive pulmonary disease in 1 patient (1.1%). Inhaled therapeutics, which are an established precipitating factor for SPM in Western countries [1-4], were not used in any of the patients in this study (Table 3).

Chest X-rays were taken in all patients, and chest computed tomography (CT) was performed in 84 patients (92.3%). Mediastinal air was observed in 44 patients (48.4%) by chest X-ray and in 100.0% by chest CT. The Macklin effect was observed in 70 patients (83.3%). Esophagography, esophagogastroduodenoscopy, and flexible bronchoscopy were per-

formed in 10 patients (11.0%), 1 patients (1.1%), and 5 patients (5.5%), respectively. Esophagography and esophagogastroduodenoscopy were performed selectively when esophageal injuries were suspected clinically, and bronchoscopy was performed in patients with a suspected bronchial injury on CT. However, the patients did not show any organ injury during further investigation.

Most patients (79, 86.8%) were admitted, whereas 12 patients (13.2%) refused admission and were discharged after close monitoring in the emergency room. For the patients who were admitted, antibiotics (first-generation cephalosporin) and nasal oxygen were administered as a conservative treatment. The patients fasted due to the possibility of esophageal injury and were allowed to initiate oral intake after showing no clinical symptoms of esophageal injury. The mean length of hospital stay was 3.0±1.6 days (range, 1–15 days). The mean hospital stay of admitted patients was 3.2±1.6 days, and that of discharged patients at the emergency room was 1 day. The patients were discharged when they had been fully evaluated and had become asymptomatic, even if there were residual radiographic findings of PM. Among the 91 patients, 2 (2.2%) were readmitted with recurrent SPM (6 and 9 months after discharge, respectively), and both patients had underlying histories of asthma.

Discussion

Pneumomediastinum has been recognized since 1,819, when Laennec reported the disease in a case caused by trauma injuries [1]. SPM was further characterized in a case series by Hamman in 1939 [1-4]. SPM is a rare disease with a reported incidence of less than 1:44,000 [1,3]. The clinical course of SPM has been regarded as benign, and its detailed clinical manifestations and proper treatment strategies have not been fully evaluated due to its rarity. To our knowledge, this study analyzed the largest number of patients with SPM to date.

The clinical presentation of SPM can often be ignored or misdiagnosed because of its vague symptoms [2,5]. Most affected patients exhibit some of the typical symptoms, and a high level of clinical suspicion is necessary for diagnosis [3]. Every patient in our series presented with one or more symptoms. Chest pain and dyspnea were the predominant symptoms in our study, which corresponds with previous reports [2-5,8]. Other frequently reported symptoms are cervical symptoms including pain, coughing, dyspnea, and dysphagia [4,8]. Therefore, the initial differential diagnoses are numerous and include pulmonary, cardiac, musculoskeletal, and esophageal etiologies [5]. Generally, clinicians can make the differential diagnosis by taking a patient's history and performing a detailed physical examination, electrocardiography, and radiographic or endoscopic studies [5].

The trigger events are mainly associated with the Valsalva maneuver (activity, cough, vomiting, shouting, and inhalation of an illicit drug) [4]. However, trigger events are not detected in many cases (30%–40%) according to previous reports [4,8], and our data also showed that more than half of the patients (52, 56.5%) had no definite triggering events. The most remarkable finding of the physical examination was subcutaneous emphysema in many studies [2,4,8]. Hamman's sign, which is generated from the crackles heard with each beat of the heart, is also a well-known auscultative sign of SPM [3,4,8]. Fever is also a common finding [7]. However, these physical findings of SPM vary among reports [2].

To make a precise diagnosis, imaging needs to be performed [2-4,8]. The air itself and an enhanced margin of mediastinal structures can be observed on chest X-ray [4]. Additionally, some papers have emphasized that when air is present between the sternum and the anterior pericardium or surrounding the pulmonary artery without a lateral film, incorrect diagnosis may occur [2,4]. Chest X-ray is generally useful for diagnosing PM, although there have been false-negative results [9]. In our data, chest X-ray was diagnostic in only 45 patients (48.9%). Chest CT is more useful than chest X-ray because thin slices are obtained and may reveal other findings such as pulmonary disease that could generate secondary PM [9]. Using chest CT, clinicians could rule out cases involving any pathologic findings in the lung such as bullae, blebbing, bronchiectasis, and tuberculosis scarring. On CT, the Macklin effect appears as linear collections of air in the bronchovascular sheaths [9]. The Macklin effect is often seen on CT in patients who have suffered blunt chest trauma, although the Macklin effect has also been reported in CT of patients with SPM [9]. Indeed, 70 patients (83.3%)

showed the Macklin effect on CT in our series. Therefore, chest CT has to be performed in all cases if SPM is suspected. Further diagnostic tools such as esophagography, esophagoduodenoscopy, or bronchoscopy can be performed when there is high suspicion of an injury of the esophagus or trachea with clinical symptoms such as fever, sweating, leukocytosis, or continuously increasing PM [8]. However, many reports did not find any clear benefit of these diagnostic modalities, and routine use of these studies should be avoided [2,4-8]; they should only be performed in cases with diagnostic doubt [2,7]. Based on our experience, chest CT could be a routine diagnostic imaging modality for PM.

Bed rest and conservative management, such as prophylactic use of analgesics and antibiotics, and limitation of oral intake, are indicated in patients with SPM [2,4,8]. Fasting and antibiotics help to prevent mediastinitis by visceral organ perforation [2,4,8], but are unnecessary for patients without a strong clinical suspicion such as severe symptoms or high levels of inflammatory markers [4,8]. Oxygen therapy, so-called 'nitrogen washout,' accelerates the disappearance of mediastinal gas by increasing the diffusion pressure of nitrogen in the interstitium [3,4]. However, several papers have reported that this intervention is also not conclusive, unlike in pneumothorax, so routine use of oxygen therapy is not recommended [2,4,8].

Many papers argue that admission and prompt evaluation [3-6,8,10,11] should be performed to allow treatment of SPM, but we consider that it could be treated without aggressive intervention or hospitalization if patients are carefully selected. These patients could be followed as outpatients. Most patients were in their teens, twenties, and thirties; had no medical history; and were safely discharged within a few days without complications. In addition, 12 patients (13.2%) who refused admission and were treated in outpatient clinics also had no complications. An aggressive diagnostic work-up and admission are required if underlying disease or organ dysfunction is strongly suspected. Because of this problem, most of the studies to date consider that admission and evaluation are necessary. However, we believe that patients with no definite clinical symptoms that suggest organ injury could be evaluated minimally and treated in outpatient clinics to save medical resources. Indeed, 50% of patients with SPM were followed as outpatients in one study, none of whom showed exacerbation or future recurrence [2]. The reason for admission is to control severe symptoms such as continuous cough, associated infections, treatment of pneumothorax, and evaluation of suspected infection. Patients who did not have complications were discharged in this study [2]. Taking into consideration the benign course of SPM, hospitalization has to be considered when the diagnosis is in question, the underlying disease needs specific treatment, or an esophageal or tracheal perforation cannot be ruled out.

This retrospective study has shown unique findings compared with previous reports. First, in terms of precipitating factors of SPM, drug abuse has been regarded as the cause of SPM; SPM can occur due to the direct toxic action of heat and the strong vasoconstrictive action of the inhaled substances [2,4]. However, no drug abusers were enrolled in our study. Second, recurrence of SPM is very rare, reportedly ranging from 0% to 1.5% [2,5-8,10]. Our recurrence rate, 2.2%, was slightly higher than that in previous reports. Two patients showed recurrence of SPM at 13 and 25 months after the initial SPM, and both of them had an underlying asthma history. Previous research on SPM recurrence reported that such patients usually have comorbidities such as gastrointestinal ulcers, diabetes mellitus, alcoholism, mental retardation, or asthma [12]. Despite the benign clinical course of SPM, patients with comorbidities have to be informed about the possibility of recurrence of SPM.

In conclusion, SPM is a rare benign disease that presents primarily in young adults who generally make an uneventful recovery. Therefore, in the absence of concomitant symptoms or severe illness requiring inpatient care, approaches that include hospitalization and aggressive treatment should be limited and individualized. But when the diagnosis is in question, or the underlying disease needs specific treatment or an esophageal or tracheal perforation cannot be ruled out, we should take into account hospital treatment. To avoid the potentially catastrophic complications of secondary pneumomediastinum, a detailed medical history and careful physical examination are required. CT could help to discriminate other causes of pneumomediastinum.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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