

Spontaneous pneumomediastinum: 41 cases

Ivan Macia^{a,*}, Juan Moya^b, Ricard Ramos^b, Ricard Morera^a, Ignacio Escobar^a,
Josep Saumench^a, Valerio Perna^a, Francisco Rivas^a

^a Department of Thoracic Surgery, Hospital Universitari de Bellvitge, Feixa Llarga s/n, 08907-L'Hospitalet de Llobregat, Barcelona, Spain

^b Department of Thoracic Surgery and Unit of Human Anatomy, Hospital Universitari de Bellvitge, School of Medicine, University of Barcelona, L'Hospitalet de Llobregat, Barcelona, Spain

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Abstract

Objective: Spontaneous pneumomediastinum is characterized by the presence of interstitial air in the mediastinum without any apparent precipitating factor. The purpose of this study is to review and discuss our experience with this condition. **Methods:** A descriptive, retrospective study of 41 cases -34 men (83%) and 7 women (17%) -treated at our hospital for spontaneous pneumomediastinum from January 1990 through June 2006. **Results:** The mean age of the patients was 21 years (range, 14–35 years). Notably, 22% of patients had a prior history of asthma. No precipitating factor was identified in 51% of cases while onset was associated with physical effort in 12%. Chest pain (85%) and dyspnea (49%) were the most common symptoms. Subcutaneous emphysema, which presented in 71% of patients, was the most common sign. Pneumomediastinum was diagnosed by plain chest radiography in all cases. In certain cases, a computed tomography scan of the chest, contrast-enhanced swallow, or bronchoscopy was performed. All patients were admitted to the hospital with good progress and no instances of morbidity or mortality. Treatment included analgesia, rest, and/or initial oxygen therapy. The mean length of hospital stay was 5 days (range, 1–9 days) with only one case of early recurrence, which was resolved satisfactorily. **Conclusions:** Spontaneous pneumomediastinum is a benign process primarily affecting young men. Despite its low incidence, spontaneous pneumomediastinum should be considered in the differential diagnosis of acute chest pain because it requires a high index of suspicion. Patients with spontaneous pneumomediastinum respond well to medical treatment, with no recurrence in the great majority of cases.

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1. Introduction

Spontaneous pneumomediastinum is defined as the presence of interstitial air in the mediastinum without any apparent precipitating factor. Cases in which the etiology of the pneumomediastinum is clear -such as pneumomediastinum associated with trauma, hollow organ perforation, iatrogenic injuries, infections, and surgery -are not considered spontaneous pneumomediastinum. Spontaneous pneumomediastinum was described by Louis Hamman in 1939, which is why it is also called Hamman's syndrome.

The pathophysiology of this condition is based on the existence of a pressure gradient between the alveoli and the lung interstitium; this pressure difference may lead to alveolar rupture and the consequent escape of air into the interstitium. Once the air is in the lung interstitium it flows towards the hilum and the mediastinum along a pressure gradient between the lung periphery and the mediastinum [1].

Spontaneous pneumomediastinum is an uncommon, benign entity primarily affecting young adults. Chest pain, dyspnea, and subcutaneous emphysema are the most common clinical manifestations. Given its low incidence, it is unsurprising to find that the only published reports of this entity are of small case series and individual case studies.

The objective of this study was to discuss our experience in the diagnosis and management of this clinical condition.

2. Materials and methods

We report a simple descriptive, retrospective study of a series of 41 clinical cases treated for spontaneous pneumomediastinum at the Hospital Universitari de Bellvitge in L'Hospitalet de Llobregat (Barcelona), Spain between January 1990 and June 2006. Patients had to fulfill the following four inclusion criteria: the presence of a clinical picture consistent with pneumomediastinum; the absence of a clearly defined triggering cause; the presence of interstitial air in the mediastinum; and, finally, the patient had to be older than 13 years of age. Exclusion criteria included

* Corresponding author. Tel.: +34 606021526; fax: +34 932607983.
E-mail address: ivanmacia@yahoo.com (I. Macia).

evidence of a clear trigger for the pneumomediastinum, such as perforation of the tracheobronchial tree or the esophagus; iatrogenic factors (manipulation of the throat or larynx); pneumomediastinum occurring after thoracic or cardiac surgery; chest wounds and injuries; infection by gas-producing germs; or any disease involving the neck or abdomen.

All patients in the study were either admitted directly or referred by other hospitals in our public health area to the emergency department at our hospital, where they were given treatment.

Patient data collection was performed according to strict protocols, with the following data collected: administrative and demographic data; predisposing factors; precipitating factors; symptoms; signs; additional diagnostic tests performed—especially imaging tests and findings; initial diagnosis upon admission; treatment administered during hospitalization and upon discharge; duration of hospitalization; and course of disease including any complications or readmissions.

We should note that *predisposing factors* were considered to be those habits or nosologic diseases from the patient's prior history that created the favorable base conditions for the emergence of spontaneous pneumomediastinum. *Precipitating factors*, on the other hand, were those events closely linked in time to the specific process of spontaneous pneumomediastinum (Table 1).

Patient data was analyzed after the data collection process had been completed for all patients. The results of this analysis are described in Section 3.

This study was approved by our institutional committee on human research. All patients provided written informed consent.

3. Results

From January 1990 through June 2006, 1,824,967 patients were treated at the emergency department at our hospital. Of these, 41 fulfilled the inclusion criteria for the study. The approximate incidence of spontaneous pneumomediastinum was, therefore, 1 in 44511 patients (22 cases per 1,000,000) treated at the emergency department. Of these 41 patients, 34 were men (82.9%) and 7 women (17.1%). The mean age was 21.3 years (range, 14–35 years) with a standard deviation (SD) of 4.8 years.

Table 1
Predisposing and precipitating factors included in the data collection protocol

Predisposing factors	Cases (%)	Precipitating factors	Cases (%)
Tobacco	34	Nausea or vomiting	9.8
Illicit drug use	9.8	Cough	7.3
Corticosteroids and other drugs	0	Sneezing	0
Interstitial lung disease	0	Defecation	0
Asthma	22	Giving birth	0
Inhalation of irritants	0	Superinfection of upper airways	7.3
		Bronchospasm	0
		Physical exercise	12
		Inflating balloons	0
		Spirometry	0
		Playing wind instruments	0
		Mountaineering	0

Table 2
Symptoms and signs

Symptoms	Cases (%)	Signs	Cases (%)
Chest pain	85	Subcutaneous emphysema	66
Dyspnea	49	of the neck	
Neck pain	44	Subcutaneous emphysema	29
Odynophagia	37	of the chest	
Cough	24	Pharyngolaryngeal	27
Crackles/clicking	12	abnormalities	
Dysphonia	12	Hamman's sign	12
Dysphagia	12		

Several predisposing factors were identified: 14 patients (34.1%) were smokers—a finding consistent with the percentage of smokers (37.5%) in Catalonia (Spain) [2], 9 patients (21.9%) had asthma, and 4 patients (9.8%) were users of illicit drugs.

In terms of precipitating factors, the absence of any specific trigger (21 cases; 51.2%) was the most common finding. The next most common factors, in order of prevalence, were physical exercise (five cases; 12.2%), vomiting (four cases; 9.8%), and cough (three cases; 7.3%). Symptoms of a superinfection of the upper airways were found in three patients (7.3%). The different precipitating factors in the remaining five cases (12.2%) were categorized together as 'miscellaneous factors' (Table 1).

The most commonly reported symptom was chest pain, which was described in 35 cases (85.4%) (Table 2); more specifically, 23 of these patients (56.1% of the series) reported centrally-located chest pain. Dyspnea (20 cases; 48.8%) and neck pain (18 cases; 43.9%) were the second and third most common symptoms, respectively. Odynophagia (15 cases; 36.6%) and cough (10 cases; 24.4%) were other common symptoms.

The mean time between the onset of the symptom and the hospital visit was 17 h and 45 min, the median was 12 h and the range was 5–72 h.

Subcutaneous emphysema was undoubtedly the most common sign, presenting -to varying degrees -in 29 patients (70.7%). Twenty-seven patients (65.8%) had subcutaneous emphysema of the neck while 12 patients (29.2%) presented with subcutaneous emphysema of the chest wall. Abnormalities associated with the pharyngolaryngeal region were described in 11 cases (26.8%). We should point out that Hamman's sign -which is pathognomonic of pneumomediastinum and described as crunching, crackling, or bubbling

sounds synchronous with the heart beat upon auscultation [3] -only presented in five cases (12.2%) (Table 2). All patients had at least one or more symptoms and/or signs.

There were two main types of diagnostic tests performed: imaging tests and laboratory tests. In terms of the imaging tests, all patients underwent plain chest radiography and the findings were consistent with pneumomediastinum in every case. In addition, 27 patients (65.8%) had radiographic signs suggestive of subcutaneous emphysema of soft tissues. Eight patients underwent plain radiography of the neck and the presence of air in the soft tissues was confirmed in all eight cases. Spontaneous pneumomediastinum was diagnosed in 32 cases (78%) after evaluation of the symptomatology and chest X-rays. If any diagnostic doubt remained -especially in cases with perforation of the esophagus and/or of the tracheo-bronchial tree -additional diagnostic tests were performed. Seven patients underwent a computed tomography (CT) scan of the chest; the findings in all cases were suggestive of pneumomediastinum and there were no signs of other diseases on the CT scan. No significant abnormalities were found in the eight patients who underwent a contrast-enhanced swallow study. Findings from the one bronchoscopy that was performed were normal.

The most noteworthy result of the laboratory tests performed (complete blood count, routine biochemistry, coagulation study, arterial blood gas analysis) was from the blood count. Of the 87.8% of patients who underwent a complete hemogram, 41.7% were found to have an elevated white blood cell count and/or neutrophilia. An electrocardiogram was performed in 24 cases. In one case, an S wave was observed in bipolar lead I with a Q wave and a negative T wave in lead III. In another case, a slight elevation of the ST segment in precordial leads was observed.

All patients with spontaneous pneumomediastinum were admitted to the hospital. The mean length of hospitalization was 5 days, with a range (SD) of 1–9 (1.76) days. Treatment included analgesia (85.4%), rest (68.3%), and initial oxygen therapy (29.3%). Additional treatments included bronchodilator (10 cases) and antibiotic (5 cases) therapy. Follow-up treatment was determined individually, based on the clinical picture and plain chest radiography (performed in 75.6% of cases). Patient progression was satisfactory as all patients showed a decrease in clinical manifestations of the condition. Radiographic signs of pneumomediastinum decreased in 21 cases (51.2%) and completely disappeared in the remaining 20 cases (48.8%). There were no significant complications, although the presence of a self-limited fever or low-grade fever in six patients should be mentioned. There was only one case of early recurrence, which responded well to conservative treatment.

4. Discussion

Spontaneous pneumomediastinum or spontaneous mediastinal emphysema was officially described for the first time by Louis Hamman in 1939, although pneumomediastinum has been known to exist since 1819, when it was reported by René Laennec [3,4]. Spontaneous pneumomediastinum is defined as the presence of interstitial air in the mediastinum without any clearly defined precipitating factor (that is, 'sine

causa'). There is some controversy as to whether pneumomediastinum associated with preexisting lung disease should be considered spontaneous pneumomediastinum since the pneumomediastinum could be attributed to the underlying disease in patients with lung disease. However, if we follow this line of reasoning, pneumomediastinum should also be excluded when it occurs in asthmatic patients. Yet, in the medical literature, such cases are not excluded.

The incidence of spontaneous pneumomediastinum has not been clearly established because the only published reports available are case studies or small case series. Newcomb and Clarke [5] reported an incidence of 1 in 29,670 emergency department presentations, a figure similar to what we observed in our series (1 in 44,511). However, the incidence of this entity is probably underestimated because it is easily ruled out when the index of diagnostic suspicion is not high; moreover, the symptoms of spontaneous pneumomediastinum are not very specific, some signs may go unnoticed, and some radiographic signs are difficult to identify.

Spontaneous pneumomediastinum primarily affects young adult males, as evidenced by the fact that more than 75% of reported cases consist of males with a mean age of 20 years [4–7]. The pathophysiology of this condition was described -based on findings from laboratory animals -by Macklin and Macklin [1] in 1944 and later revised by Maunder et al. [8].

Pneumomediastinum can be produced, in general, by three different mechanisms, as follows: (1) gas-producing microorganisms present in an infection of the mediastinum or adjacent areas; (2) rupture (whether traumatic or not) of the cutaneous or mucosal barriers-especially perforation of the esophagus or tracheobronchial tree, allowing air to enter the mediastinum; and (3) the existence of a decreasing pressure gradient between the alveoli and the lung interstitium that can result in alveolar rupture.

This pressure gradient can be produced by increasing intraalveolar pressure -as occurs in techniques such as the Valsalva maneuver, or by decreasing pleural pressure -which happens when the Mueller maneuver is performed in bronchial asthma; or by decreasing interstitial pressure -as occurs in intense work of breathing and vasoconstriction. Alveolocapillary membrane abnormalities and interstitial lung diseases can also favor the development of a rupture [9]. Alveolar rupture leads to the accumulation of air in the interstitium that circulates centripetally through the venous sheaths to the hilum and mediastinum (Macklin effect) [1]. This occurs because the pressure in the mediastinum is lower than that of the lung periphery. Once in the mediastinum, the air decompresses into the cervical space, soft tissues, or even the retroperitoneal space. This third mechanism is the pathophysiological basis of spontaneous pneumomediastinum.

Both predisposing and precipitating factors of spontaneous pneumomediastinum have been described in the medical literature; however, no distinction has been made between them, despite the fact that they are not the same. Asthma has been described in 8–39% of cases [4,5,8,10,11] and is one of the most commonly reported factors. Illicit drug use has traditionally been associated with spontaneous pneumomediastinum [4,12]. Other associated factors include those that provoke a Valsalva maneuver: coughing, sneezing,

defecation, giving birth, nausea, and vomiting [5,8,11]. The symptoms of a superinfection of the upper airways reported in our study may reflect a coughing or sneezing mechanism that was unnoticed by the patient. Finally, intense breathing work or exercise, which take place during respiratory function tests or intense physical exertion, have also been associated with spontaneous pneumomediastinum [4].

The most common symptoms described in the literature are chest pain, dyspnea, and neck pain or discomfort. Chest pain is the most commonly reported symptom in most series [4–7,10,11] and typically presents as an acute, retrosternal, pleuritic pain that may radiate to the neck, back, or shoulders. Other, less common symptoms include odynophagia, cough, dysphonia, back pain, dysphagia, or abdominal pain. The general state of patients presenting with spontaneous pneumomediastinum is usually good; patients are usually hemodynamically stable and eupneic. Subcutaneous emphysema -especially of the neck- has a reported prevalence ranging from 40% to 100% and is the most commonly reported sign [4–7,11]. The reported prevalence of Hamman's sign is highly variable, ranging from 0% to 100% [5–7,10,13]. In the most recently published case series, the prevalence of Hamman's sign was less than that of previously reported series; a relative decrease was also noted in our series, in which 12% of patients presented with Hamman's sign.

In our series, the three most common clinical manifestations of spontaneous pneumomediastinum were chest pain, dyspnea, and subcutaneous emphysema of the neck. These findings are consistent with those reported by other studies.

Spontaneous pneumomediastinum is an acute disease, and this is reflected in the mean interval time between the onset of the symptom and the hospital visit, which is 17 h and 45 min. This fact shows the necessity of an agile examination and performance of diagnostic tests.

Diagnosis was confirmed by posteroanterior and lateral chest radiography. A lateral view is necessary because studies have found that up to 50% of all cases may remain undiagnosed if only a posteroanterior radiograph is taken. In cases of pneumomediastinum with small quantities of air, the only sign is a radiolucent band (hyperlucency) in the retrosternal area [14]. However, pneumomediastinum has multiple radiographic signs [15], including a band of hyperlucency parallel to the left side of the cardiac silhouette with a fine radiopaque line indicating the elevated mediastinal pleura; radiolucent lines in the mediastinum extending towards the neck; and air surrounding mediastinal structures such as the aorta, trachea, esophagus or thymus gland. The presence of subcutaneous emphysema of soft tissues (especially in the neck and less frequently in the chest) is associated with the aforementioned signs in a high percentage of patients (Figs. 1 and 2). As in other reported series, we found radiographic signs suggestive of pneumomediastinum in 100% of the cases in our series [4,6,7]. However, as Kaneki et al. [16] point out, up to 30% of patients with spontaneous pneumomediastinum present with a normal radiograph, which is why those authors recommend that a CT scan of the chest (Fig. 3) be performed. The chest CT scan is considered the gold standard of imaging tests, capable of detecting pneumomediastinum even in patients with small amounts of mediastinal air or when the Macklin

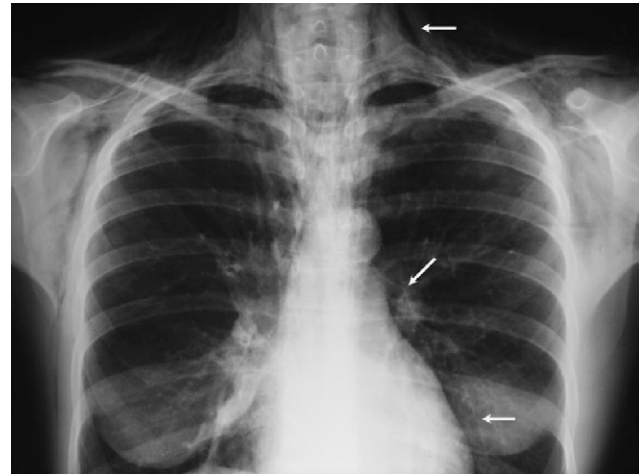


Fig. 1. Example of a posteroanterior plain chest radiograph showing signs of pneumomediastinum and subcutaneous emphysema in soft tissues (arrows).

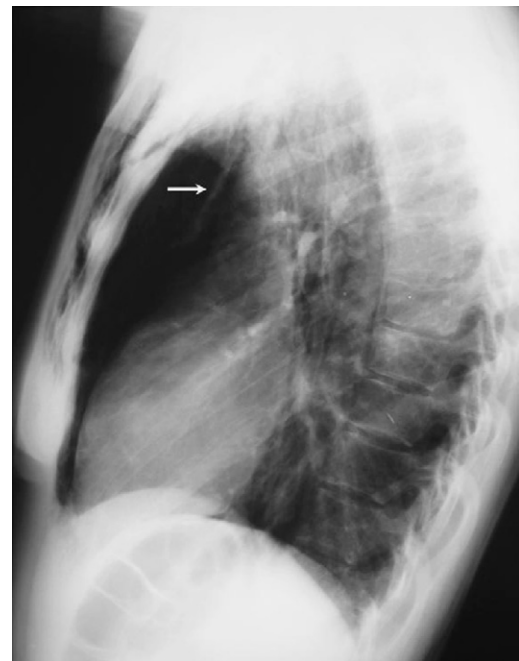


Fig. 2. Example of a lateral chest radiograph showing a radiolucent band previous to great vessels (arrow), sign of pneumomediastinum.

effect is present [17]. Considering these results, chest CT scans should be reserved for those cases in which the diagnosis is unclear.

The differential diagnosis of spontaneous pneumomediastinum must include several other diseases, some of which - musculoskeletal disorders, acute coronary syndrome, pericarditis, pneumothorax, pulmonary embolism, and, especially, tracheobronchial tree rupture and Boerhaave's syndrome -are potentially dangerous [4].

Boerhaave's syndrome is accompanied by retrosternal chest pain and subcutaneous emphysema of the neck, and usually affects patients after vomiting. Chest radiography can detect pneumomediastinum, subcutaneous emphysema of soft tissues, pleural effusion, and, at times, pneumothorax. The blood count of patients with Boerhaave's

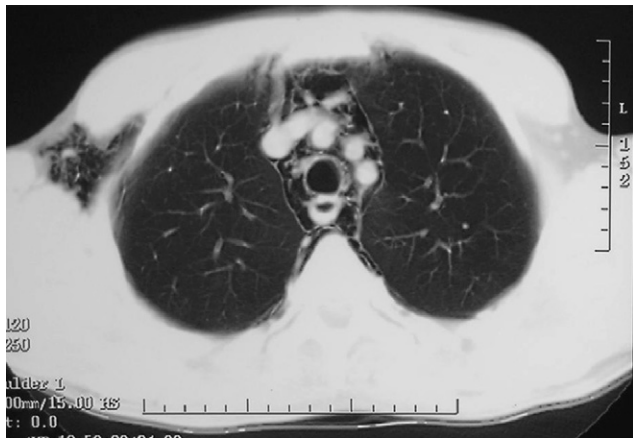


Fig. 3. Example of computed tomography scan of the chest showing the presence of interstitial air surrounding the structures within the mediastinal pleura.

syndrome reveals a marked increase in white blood cells. Clearly, Boerhaave's syndrome and spontaneous pneumomediastinum share some common features. In case of doubt (general malaise, low blood pressure, tachycardia, fever, pleural effusion, coagulation abnormalities), additional tests -such as contrast-enhanced CT scan of the chest or contrast-enhanced swallow -become more important. Bronchoscopy is the definitive test in cases with a suspected tracheobronchial tree rupture.

There are three main approaches to the treatment of spontaneous pneumomediastinum: rest, oxygen therapy, and analgesia. Patients respond well to this treatment: clinical manifestations resolve and radiographic signs of the condition diminish [4–7,10]. Most healthcare clinics recommend that patients be admitted to the hospital for 2–5 days of observance, similar to what occurred in our series [5,7,13]. Significant complications are virtually nonexistent and most published series have not reported any cases of recurrence, although Abolnik et al. [6] and Gerazounis et al. [18] reported one case of late recurrence and we had one case of early recurrence in our series. In all cases, patient progress was satisfactory. Given the low incidence of recurrence, outpatient follow-up is not necessary.

We can conclude that spontaneous pneumomediastinum is an uncommon and benign process primarily affecting young adult men. Its pathophysiology is based on the production of a pressure gradient between the alveoli and the interstitium, leading to alveolar rupture. The traditional clinical triad includes chest pain, dyspnea, and subcutaneous emphysema of the neck. The diagnostic test for spontaneous pneumomediastinum is plain chest radiography with both posteroanterior and lateral views. Other tests should be reserved for selected cases, especially to rule out hollow organ perforation. Patients with spontaneous pneumomediastinum respond well to medical treatment, with no recurrence in most cases. In our study -and in most other published studies -patients were admitted for in-hospital observation; however, in certain selected cases, domiciliary observation may be sufficient.

In conclusion, despite its low frequency, spontaneous pneumomediastinum should be considered in the differential diagnosis of acute chest pain because it requires a high index of suspicion and because the treatment protocol differs substantially from that of many other processes with similar clinical features.

As larger and larger case series are reported in the future, we will be able to more clearly establish both the incidence of spontaneous pneumomediastinum and the validity of current treatment protocols.

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