



# Spontaneous Pneumomediastinum: A Case Series

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## Abstract

**Introduction:** Spontaneous Pneumomediastinum (SPM) is a benign condition defined as the presence of air in the mediastinum. Most cases are discovered incidentally. Symptoms may include chest pain, throat pain, dysphonia, dysphagia and dyspnea. Clinical course is generally uneventful, and most patients are managed conservatively.

**Presentation of Cases:** We present 2 cases of young, fit male patients. The first patient presented with throat pain for 2 days. Clinical examination was insignificant. Radiographic investigations demonstrated evidence of pneumomediastinum with subcutaneous emphysema. The second patient was brought with 1-h history of agitation. On examination chest wall emphysema was demonstrated. Computed Tomography (CT) scan demonstrated pneumomediastinum, pneumoperitoneum and pneumoretroperitoneum. Both patients were treated conservatively and discharged well within days.

**Conclusion:** Cases of SPM are not uncommon. CT scan and endoscopy are the cornerstone of diagnosis. Management is primarily conservative. More studies are required to accurately pin down the true incidence rate of SPM and the ideal management plan.

## Introduction

Pneumomediastinum is defined as the presence of air in the mediastinum. This can be caused secondary to trauma (iatrogenic injury or external trauma), infections, predisposing lung disease or surgery. The source of air in these cases can be the esophagus, lungs or bronchial tree. Primary pneumomediastinum, also known as Spontaneous Pneumomediastinum (SPM), is an uncommon, self-limiting, benign condition that predominantly affects young males (13 to 35 years) and pregnant females [1]. In contrast to secondary pneumomediastinum, no identifiable cause can be pinned down in spontaneous pneumomediastinum. Spontaneous pneumomediastinum was first described in 1939 by Louis Hamman, hence its alternative name: Hamman Syndrome.

The pathophysiology of SPM was demonstrated by Macklin et al. in 1944 [2]. Rupture along the alveolar tree leads to the release of air from the alveolar spaces to the pulmonary interstitium. This occurs due to the pressure gradient between the intra-alveolar spaces and the lung parenchyma. The release of air in a centripetal manner causes dissection of the pulmonary interstitium, bronchovascular sheaths and progresses towards the pulmonary hila and the mediastinum down its pressure gradient. This was later revised by Maunder et al. in 1984.

Patients commonly present with retrosternal chest pain that is acute and pleuritic in nature and may radiate to the neck, back or shoulders. Chest pain is the most commonly reported symptom in most studies. Other symptoms include dysphagia, dysphonia and dyspnea. Generally, the patients are in a good condition at presentation. They are hemodynamically stable and do not suffer from acute dyspnea. Chest pain, dyspnea and subcutaneous emphysema are demonstrated clinically. Other findings reported include the absence of cardiac dullness to percussion and Hamman's sign. Hamman's sign is a crunching rasping sound heard over the precordium synchronous with heart sounds. It is a result of the heart beating against air-filled tissues.

This report describes 2 cases of SPM admitted and managed in Sheikh Khalifa Medical City in Ajman, United Arab Emirates, during the period between 2016 to 2018.

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**Figure 1:** CT scan with contrast lung window (Coronal & sagittal sections) revealed pneumomediastinum (black arrow) with air extended up causes neck & chest wall surgical emphysema (white arrow).

## Case Presentation

### Patient 1

A 19-year-old previously healthy male came presenting to the outpatient department with complaints of throat pain for a duration of 2 days. The pain is localized to the throat with associated neck pain but otherwise does not radiate anywhere else. It is not associated with cough, fever, chest pain, or shortness of breath. There is no recent history of abdominal pain, vomiting or retching, no history of anxiety, depression or eating disorders. No significant history of chronic lung diseases or structural defects. History is negative for any trauma, recent medical intervention, surgery or endoscopic procedure. The patient is well built, active and exercises regularly.

On examination, the patient looked comfortable, vital signs were normal; the neck was tender on the left side with a palpable left thyroid lobe. Chest examination revealed subcutaneous crepitus and clear bilateral air entry. Chest X-Ray showed pneumomediastinum with air extending upwards into the subcutaneous tissue at the base of the neck. No evidence of pneumothorax was seen. Both lung fields were clear. Cardiac shadow was normal.

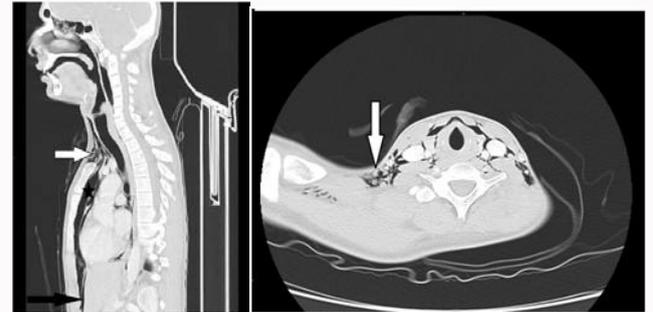
Computed Tomography (CT) scan with contrast revealed pneumomediastinum (black arrow) with subcutaneous emphysema seen along the mediastinum down to the diaphragm. Pneumopericardium can also be seen along the pericardial sac. Air is seen extending upwards and causing neck & chest wall emphysema (white arrow).

Emphysema seen around the neck vessels reaching posteriorly below the scapula bilaterally. There is no evidence of pneumothorax or pleural effusion. Normal appearance of the other mediastinal structures. The patient was admitted and treated conservatively and was later discharged after an uneventful stay. Prior to discharge, chest X-ray was done and revealed unremarkable findings Figure 1.

### Patient 2

A 22-year-old previously healthy male prisoner was brought to the emergency department with history of agitation for 1 h. He had no recent complaint of dyspnea, dysphonia, cough, chest pain, or fever. Dysphagia, abdominal pain, vomiting and retching were negative too. His past medical history was unremarkable for chronic lung disease, trauma, recent medical intervention, surgery or endoscopic procedure.

On examination, he was disoriented. Neck examination showed palpable subcutaneous emphysema. Other elements of physical



**Figure 2:** CT scan with contrast lung window (sagittal & axial sections) revealed pneumomediastinum (black star) with air extended up causes neck & chest wall surgical emphysema (white arrow) and through the diaphragm causes pneumoperitoneum (black arrow) and pneumoretroperitoneum.

examination were unremarkable.

CT scan with IV contrast was done to exclude any possibility of trauma. Extensive neck and chest wall emphysema was manifested as bilateral parapharyngeal air (white arrow). Extensive pneumomediastinum was present too, with air extended into both lungs (white star) and diaphragm causing pneumoperitoneum (black arrow) and pneumoretroperitoneum.

CT scan with oral contrast was then done and revealed no foreign body or other definitive cause of generalized emphysema. An Esophagogastroduodenoscopy (EGD) was done and it was unremarkable. A rare anatomical anomaly known as Os odontoideum (a fragmented odontoid process separated from the main body of the axis was incidentally discovered by the CT.

The patient was managed conservatively, and psychiatric evaluation was performed. His condition was diagnosed as Generalized Anxiety Disorder (GAD) and was treated accordingly. The period of his stay in the hospital was uneventful Figure 2.

## Discussion

Clear reports on the incidence of SPM are not available. This is mainly because the only reports available about the incidence are case studies or small case series. Not to mention the underestimation of the actual incidence of SPM since it can be easily missed if there is a low index of suspicion. Many patients are diagnosed incidentally as seen in patient 2 of this article. Moreover, radiological signs can be quite challenging to recognize, and thus, many cases remain undiagnosed. Available reports estimate an incidence of 1 in 29,670 as reported by Newcomb and Clarke [3]. Others indicate an incidence between 1:7000 to 1:45000 [4,5].

More than 70% of reported cases appear to consist of males with an age range of 13-35 years and a mean age of 20. This is similar to the patients reported in this case series. Pregnant females are also commonly affected by SPM. Reports show that frequent exercise, especially weightlifting, can be a precipitating factor for the development of SPM. Patient 1 of this case series was well built and exercised regularly. However, weightlifting is rarely reported as the cause of SPM. Interestingly, a recent study demonstrated the development of SPM following electronic cigarette use [6].

The pathophysiology of SPM was first described using animal models by Macklin in 1944. His studies show that hyperinflation of the alveoli can lead to their rupture causing leakage of air from the alveolar spaces to the interstitium and migration of air towards the lung

hila and mediastinum leading to SPM [7]. Once in the mediastinum air can migrate and travel across tissue planes upwards towards the neck and face, downwards towards the abdomen and laterally to the limbs causing subcutaneous emphysema or pneumoperitoneum as in patient 2 of this case series. Three mechanisms for the development of SPM as described by Macia include rupture of the aero-digestive tracts due to traumatic or non-traumatic causes, existence of a pressure gradient between the alveoli and lung parenchyma or infections due to gas formic microorganisms.

Several studies have reported a myriad of precipitating factors such as retching, severe vomiting as seen in Diabetic ketoacidosis [8,9], inhalational drug abuse such as cocaine use [10,11] and barotrauma. Barotrauma is seen with instrumentation, mechanical ventilation and Valsalva maneuver which occurs when the patient strains such as during labor. Rarer precipitating factors include inhalation of irritating vapors [12] and helium gas from party balloons [13]. However, none of those precipitating factors were reported by our patients.

In contrast to our patients who were non-smokers and non-asthmatics, a study conducted by Macia showed that 34% of patients included in their study had tobacco smoking as their predisposing factor, 22% were asthmatic and 9.8% engaged in illicit drug use. Asthma has been described in 8% to 39% of cases and is one of the most commonly reported factors to lead to the development of SPM.

The most common presenting symptom is chest pain. Other symptoms less often reported by the patients include neck pain, cough, dysphagia, dysphonia, dyspnea, and light-headedness. Nasal voice is another rare clinical presentation reported in literature. Throat pain and neck pain were reported in patient 1 of this case series, while patient 2 was asymptomatic and findings of SPM were incidental. Differential diagnoses include acute coronary syndrome, pericarditis, pneumothorax, pulmonary embolism, and Boerhaave syndrome. Once excluded by various investigations and by clinical elimination, a diagnosis of SPM can be made.

Some studies suggest a Complete Blood Count (CBC) and CT chest with IV contrast for all patients even if Chest X-Ray (CXR) showed clear signs of pneumomediastinum. In contrast, Iyer suggest that CXR appeared to be relatively sensitive at diagnosis SPM. Al-Muffarej do not recommend all patients to go for contrast swallow studies. They suggest a contrast swallow study for patients with emesis, dysphagia, gastrointestinal disease, trauma, hemodynamic instability, fever, leukocytosis, pleural effusion or pneumoperitoneum. Diagnostic endoscopy should also be performed in patients where esophageal perforation is highly suspected, if positive, surgical intervention is indicated. The need for additional investigations is only warranted when there is a clear question as to what the cause of the mediastinal emphysema is in the absence of a specific clinical history or uncomplicated radiographic images [14]. Both patients in this case series did a CT scan and demonstrated radiographic evidence of SPM.

SPM is a benign condition that is often managed by reassurance and simple measures including admitting the patients for monitoring, avoiding triggers, bed rest and oxygen. Intensive care is indicated in hemodynamic instability or in cases of high suspicion of esophageal injury.

Complications are rare but could include hypertensive pneumomediastinum which happens due to cardiac and large vessel

compression causing decreased venous return from the body and thus hemodynamic instability and respiratory compromise [15]. Mediastinitis is also a serious complication. However, there are no reports of mortality directly attributed to SPM in the literature at the time of writing this article. Our patients did not suffer from any complications during their period of admission in the hospital.

Recurrence of SPM is very rare. It can however recur in case of repeated exposure to the trigger such as in continued use of inhaled drugs or exposure to inhaled irritants or in cases of unavoidable exposure such as asthma.

## Conclusion

Cases of SPM are not uncommon. CT scan and endoscopy are the current cornerstone of diagnosis. Management is primarily supportive and conservative. With more studies, we can more accurately pin down the true incidence rates of SPM. It will also allow us to assess the current treatment protocols and evaluate the efficacy of the current management plans and diagnostic workup as well as evaluate the long-term sequelae of conservative management of such cases. This will be of great use for the diagnosis of SPM and exclusion of more severe differential diagnoses that warrant immediate management.

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