

Case Series

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Spontaneous pneumomediastinum: case series and literature review

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ABSTRACT

Spontaneous pneumomediastinum (SPM) is a condition characterised by presence of air in the mediastinum that is not iatrogenic or secondary to trauma. We conducted a retrospective review of 4 cases, treated at our hospital for SPM in 2023. The mean age of the patients was 35 years (range, 28-58 years), with 3 male (75%). Only one of the patients (25%) had precipitating projectile vomiting followed by hematemesis. Chest pain was by far the most common symptom (75%). One patient presented with haematemesis, raised inflammatory markers and fever. Pneumomediastinum was diagnosed by plain chest radiography in all cases. In all cases a computed tomography (CT) scan of the chest with on table contrast was performed with half of the patients needing fluoroscopy swallow later in the admission. Half of the patients were treated for suspected oesophageal perforation and received antibiotics and kept nil by mouth, the other two patients only required supportive care. The mean length of hospital stay was 3.4 days (range, 1.2-4.7 days). SPM is a benign process primarily affecting young otherwise healthy males. There is a growing body of evidence to suggest that CT of the chest with on table contrast should be reserved for patients presenting with red flags such as abdominal pain, hematemesis, fever and raised inflammatory markers.

Keywords: Spontaneous pneumomediastinum, Mediastinal emphysema, SPM, Hamman's syndrome

INTRODUCTION

Spontaneous pneumomediastinum (SPM) is a rare medical condition where air gets trapped in the mediastinum without apparent causes like trauma, bacterial infections, or medical procedures. The incidence of this condition ranges from 1 in 800 to 1 in 42,000.¹ The most common causes of this condition are activities that increase the pressure inside the chest, like coughing, vomiting, defecating or heavy lifting.¹ The most common symptoms of SPM are chest pain, difficulty breathing and subcutaneous emphysema.² This study aims to describe four cases of SPM that were treated at a level 1 trauma centre in Sydney, Australia, in the year 2023. We conducted a comprehensive review of current

recommendations by evaluating published literature on the subject.

CASE SERIES

The study conforms to the provisions of the declaration of Helsinki (as revised in 2013). The informed consents were obtained from all four patients.

The files of four patients diagnosed with SPM were reviewed. The demographic characteristics and clinical and laboratory data of the patients included in the study were noted separately for each. Demographic parameters included age, gender, smoking status, and comorbidity. Patients' body temperature, mean arterial blood pressure were measured. Symptoms such as cough and shortness

of breath were recorded at admission. Finally, the laboratory values collected include leucocyte count and C-reactive protein (CRP). Chest radiographs and low-dose thorax CT images of all patients were examined. The clinical, laboratory, and demographic characteristics of the patients at the first admission were tabulated and presented (Table 1). As we have only identified four patient the detailed case description can be found below. The mean age of the patients was 35 years (range, 28-58 years), with 3 male (75%).

Only one of the patients (25%) had precipitating projectile vomiting followed by hematemesis. Chest pain was by far the most common symptom (75%). One patient presented with haematemesis, raised inflammatory markers and fever. No patients exhibited subcutaneous emphysema or Hamman's sign. Pneumomediastinum was diagnosed by plain chest radiography in all cases, however, was missed by a reporting radiologist in one of the patients. In all cases a CT scan of the chest with on-table contrast was performed with half of the patients requiring fluoroscopy swallow later in the admission. All patients were admitted to the hospital with good progress and no instances of morbidity or mortality. Half of the patients were treated for suspected oesophageal perforation and received antibiotics and kept nil by mouth, the other two patients only required supportive care. The mean length of hospital stay was 3.4 days (range, 1.2-4.7 days).

Table 1: Summarization of cases.

Characteristics	Count	Percentages (%)
Sex		
Male	3	75
Female	1	25
Race		
Caucasian	2	50
Asian	1	25
Indigenous Australian	1	25
Age (in years)		
Mean age	35	
Smoking status		
Non-smoker	3	75
Vaping	1	25
Symptoms		
Chest pain	3	75
Vomiting	1	25
Imaging findings		
Pneumomediastinum on X-ray chest	4	100
Pneumomediastinum on CT chest	3	75
Extraluminal contrast on CT/fluoroscopic oesophagogram *	1	25

*Case 1 had equivocal CT Oesophagogram with extraluminal contrast, but no oesophageal defect identified and no contrast leak on Fluoroscopic Oesophagogram.

Case 1

A 28-year-old indigenous Australian female presented to the emergency department (ED) following a seizure with a head strike while at a concert. She did not have chest pain, shortness of breath or respiratory symptoms. Admission blood work was unremarkable, with a leucocyte count of $5.5 \times 10^9/l$ and a CRP of 5.0 mg/l. She was afebrile, and her vital signs were stable. CT scan of the brain and C spine was ordered as a part of a head injury workup, which revealed incidental subcutaneous emphysema in the lower neck. Further workup included a CT chest that confirmed the presence of a small amount of air in the mediastinum without features of mediastinitis or oesophageal injury. Upon further investigation, it was revealed that the patient presented to another hospital three days prior with a complaint of sudden onset of chest pain. She had a chest X-ray taken that was reported as normal at that time (Figure 1A and B). Chest X-ray was re-evaluated at our facility and was judged to have signs of pneumomediastinum (PM). The patient underwent the CT with on-table contrast to exclude an oesophageal injury. The test returned equivocal results with suspicion of anterior and posterior contrast leak without evidence of oesophageal defect. The patient was kept nil by mouth and had intravenous antibiotics until she had a contrast swallow study. The fluoroscopy did not demonstrate contrast extravasation. During the admission, her inflammatory markers and vital signs remained stable, and she was discharged after a neurological review for seizure management.

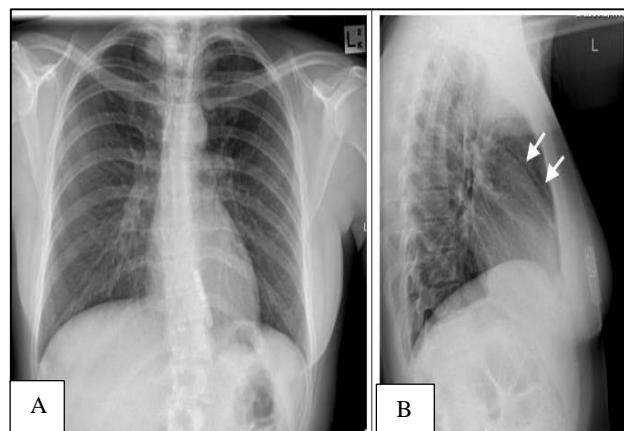


Figure 1 (A and B): Posterior-anterior (PA) erect and lateral chest X-ray projections. While PA projection appears to have evidence of pneumomediastinum, lucency around heart border is evident of the lateral projection (white arrows).

Case 2

A previously healthy 59-year-old man presented to the ED with complaints of oedema and pain in the right arm extending to the mid-arm and outpatient ultrasound (US) report demonstrating occlusive thrombus in the right axillary and subclavian veins. He is asthmatic and has no

other past medical history. He abstained from food and water for seven days as a part of the fasting routine. He did not complain of chest pain, shortness of breath or cough. He had no evidence of subcutaneous emphysema. The bloodwork was unremarkable, with a leucocyte count of $8.5 \times 10^9/l$. The CT venogram was performed for further evaluation of the occlusion. Imaging revealed small-volume PM and emphysematous changes in the lungs. There was no evidence of oesophageal or bronchial injury or mediastinitis. A contrast study was recommended by radiology consultant to exclude Boerhaave's syndrome. The neck and chest CT with on-table contrast did not demonstrate contrast leak or signs of oesophageal rupture. He had progress chest X-ray performed 2 days later which had stable appearance of PM (Figure 2). He underwent urokinase treatment for occlusive thrombus and was discharged on day 4 with vascular follow-up.

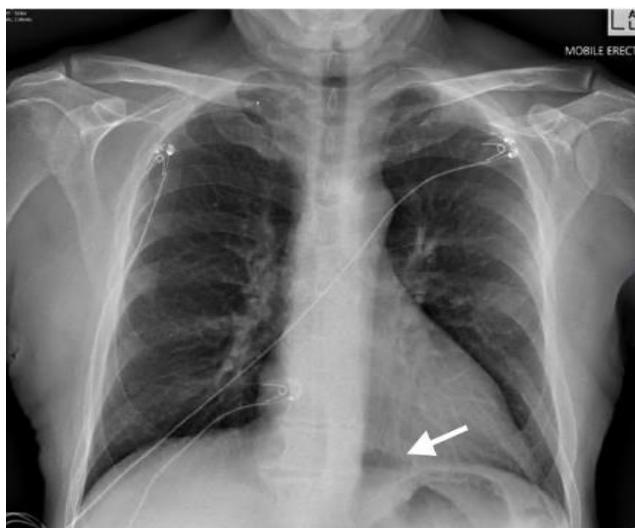


Figure 2: Anterior-posterior (AP) projection with evidence of Naclerio V sign (white arrow).

Case 3

A 29-year-old man presented to the ED with a 3-day history of chest pain, nausea and vomiting. He noticed coffee ground vomit, which prompted a presentation to the hospital. His past medical history included a recent binge episode. On arrival, he was febrile to 37.9, his blood pressure was 143/84, and his heart rate was 87. He had marked tenderness in the epigastrium with guarding and focal rigidity. His leucocyte count was $18.1 \times 10^9/l$, CRP was $<0.3 \text{ mg/l}$. Chest X-ray revealed evidence of PM with trace of lucency at the right heart border (Figure 3). CT chest demonstrated diffusely thickened mid and distal oesophagus and extensive PM without features of mediastinitis. He was treated with intravenous antibiotics, intravenous proton pump inhibitors and analgesia. The patient underwent a fluoroscopy swallow study that did not indicate oesophageal perforation. His vital signs remained stable during hospital admission, and inflammatory markers normalized. His condition improved significantly, and he was discharged on day 4 of admission.

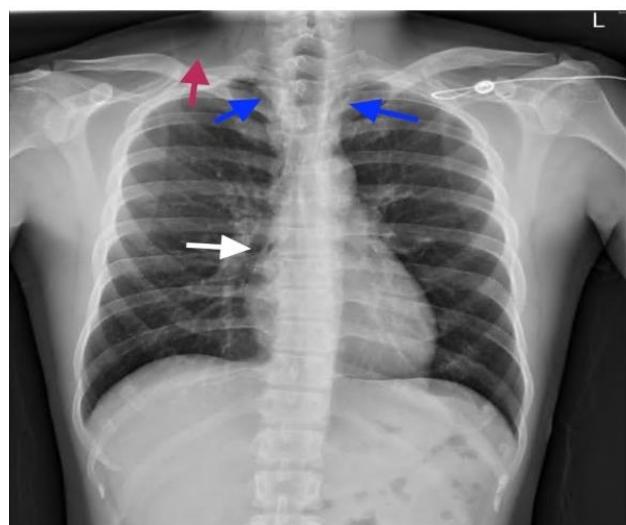


Figure 3: PA projection chest X-ray. Evidence of lucency at right cardiac border (white arrow), air in the paratracheal regions (blue arrows) and in the cervical region (red arrow).

Case 4

A 29-year-old man presented to the ED with a complaint of pleuritic chest pain that radiated to the right scapula. His vital signs were stable; he was afebrile. He had no significant past medical history aside from occasional vaping 2-3 times a week. He had normal inflammatory markers with leucocyte count of $5.0 \times 10^9/l$ and CRP of 6.5 mg/l . There was evidence of PM on a chest X-ray with lucency at the right heart border, and air extending into his neck. (Figure 4) CT chest with on-table contrast revealed air in the prevascular and post-vascular spaces extending from the diaphragm superiorly into the neck (including both carotid space and visceral and retropharyngeal spaces) and into the left supraclavicular region. There was no extraluminal contrast to suggest perforation. His condition remained stable, and pain was well controlled with oral analgesia. Patient was discharged following day with advice to cease vaping.

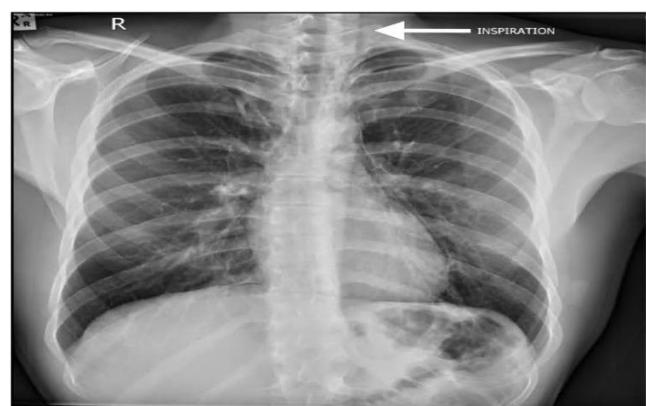


Figure 4: PA projection chest X-ray. Evidence of air in the air extending into the neck along the carotid space (white arrow).

DISCUSSION

Pneumomediastinum (PM) is a condition of the presence of air in the mediastinum. René Laennec was the first clinical to describe a case of traumatic PM in 1819.¹ An American physician, Louis Hamman, first described a case of SPM in a series of cases presented in the second Henry Sewall Lecture at Johns Hopkins medical school in 1939.¹ He mentioned a "crunching sound" when auscultating chest of an otherwise healthy 51-year-old man who presented with crushing chest pain that resolved spontaneously. The condition bears his name and is frequently referred to as "Hamman's syndrome".

SPM is rare, with a diagnosis rate of approximately 1 in 44,500 emergency attendances or 1 in 100,000 natural births. It appears to be more common in children, with reported rates ranging from 1 in 800 to 1 in 15,500. Some studies indicate an incidence of 1 in 25,000 among individuals aged 5-34 years.¹ However, many authors suggest that SPM might be more prevalent than initially thought due to underdiagnosis.¹ This often occurs as patients may not seek medical attention, and the condition might not be evident on chest radiographs, leading to symptoms being misattributed to musculoskeletal pain or other minor causes. The majority of affected individuals are male, comprising 76% of cases.¹ Some authors suggest that patients with asthma, chronic obstructive pulmonary disease, interstitial lung disease, and those who inhale tobacco or marijuana may be at an increased risk of developing PM. Stack et al reported a 0.3% incidence of SPM associated with asthma amongst patients presenting to their institution over ten years.^{1,2} Precipitating events include vomiting, coughing, upper respiratory infections, defecation, labour, physical exercise, respiratory distress syndrome, use of air instruments, and convulsions. The most common symptom of SPM is chest pain and shortness of breath. A case report by Pratibha et al describes stridor as a presenting complaint in a patient with SPM.³

The pathophysiology of PM was first described by a Canadian pulmonologist, Charles Macklin.¹ Macklin's effect explains the process of PM in cases of blunt chest trauma. The pathophysiology of PM can be explained by the difference in pressure between the alveoli and lung interstitium. Macklin proposed that alveolar ruptures due to blunt chest trauma cause air dissection along bronchovascular sheaths and interstitial emphysema, which then spreads into the mediastinum. This process can also occur in cases of SPM, where sudden increase in intrathoracic pressure due to a precipitating event (such as the Valsalva manoeuvre) can cause air to escape.^{2,3} Once the air is in the lung interstitium, it moves towards the hilum and the mediastinum along pressure gradient between the lung periphery and mediastinum. If the pressure gradient remains constant, air will follow vascular sheaths towards the mediastinum and extend outwards to the sub-pleural space. Once in mediastinum, the air may escape into the pleural spaces, neck, and

trunk, resulting in subcutaneous emphysema. Alternatively, it may travel into the retroperitoneal space, which functions essentially as a pressure release mechanism.⁴ However, if the pressure in the mediastinum continues to increase, it can lead to physiological effects resembling pericardial tamponade. This results in compression of the great vessels, which can compromise right heart filling and eventually lead to hypotension and cardiac arrest. Early clinical signs may include pulses paradoxus and electrical alternans.⁵ Anterior SPM is more common and is associated with benign course.

It is essential to acknowledge that the trachea, central bronchi, and peritoneal cavity can be a source of PM. Harris et al described the presence of PM following a laparoscopic anti-reflux surgery.⁶ In their study, nearly half (47%) of patients had evidence of extra-abdominal air on chest radiography, and 86% had CT evidence of PM. Farah and Makhoul described a case of perforated sigmoid cancer presenting as PM.⁷ Several other authors reported similar cases of the perforated colon presenting as chest pain with CT evidence of SPM.¹ It is crucial to recognize that the air may come from oesophagus. Out of all oesophageal perforations, spontaneous perforation (including Boerhaave's syndrome) represents between 8% and 33% of all oesophageal perforations, while traumatic and malignant perforations account for 17% of cases.⁷ In the healthy oesophagus, perforation usually occurs in areas of natural anatomical weakness, such as Killian's triangle, the cricopharyngeal region and the esophagogastric junction. Descending necrotizing mediastinitis (DNM) requires special considerations. The infection usually originates from peritonsillar/retropharyngeal and odontogenic source. PM can be observed in 64% of patients with DNM, according to the single-centre review done in Peking union medical college hospital, China.⁹ The presence of fever, raised leukocyte count, and C reactive protein should prompt the investigation as this increases suspicion of mediastinitis.

Susai and Banks summarised X-ray findings in their review paper.¹ One of the most common signs is lucent streaks or bubbles of air that outline mediastinum. However, there are several other radiological signs that may indicate the presence of air in mediastinum. The extra-pleural air sign was first described by Lillard and Allen in 1965.¹⁰ It is defined as the presence of gas between the parietal pleura and the diaphragm. A lateral chest radiograph can reveal a radiolucent area surrounding the right pulmonary artery when viewed on a lateral chest radiograph. It is also called the "Ring around the artery".¹ A thymic sail sign can be appreciated in infants.¹¹ Vanden and Popik described a "spinnaker-sail" sign, which represents the thymic lobes that are shifted upwards, resembling a full sail.¹² Another X-ray evidence is the continuous diaphragm sign, which was described by Levin in 1973.¹ Free air is present between the pericardium and diaphragm, causing the central parts of the diaphragm to become apparent. The Naclerio V sign is often associated

with oesophageal rupture.¹¹ It manifests as a V-shaped air collection, with one limb delineated by mediastinal gas along the left lower lateral mediastinal border and the other limb formed by gas between the parietal pleura and the medial left hemidiaphragm. A potential radiological indicator of tension PM is an earth-heart sign.¹¹ This occurs when the cardiac silhouette takes on the appearance of an oblate sphere due to an enlargement in the transverse cardiac diameter and a reduction in the vertical cardiac diameter. The reason for this is the collapsed and constrained filling of the cardiac chambers. According to Esayag et al plain chest X-ray was sensitive in 92% of patients with PM.¹³ Similar findings were reported by Wong et al.¹⁴ In their study, lateral neck soft-tissue radiographs revealed abnormal air collections in 9 out of 10 patients who had normal chest radiographs. The researchers propose that incorporating lateral neck radiographs could aid in detecting certain patients with SPM who might otherwise have normal findings on chest radiographs.

Cross-sectional CT imaging remains one of the most used modalities to investigate patients with PM further. It aids in two main diagnostic difficulties: identifying patients with false-negative chest X-rays and differentiating SPM from DNM, oesophageal or tracheal perforation of intrabdominal pathology. Okada et al described a series of 33 patients diagnosed with SPM; chest CT scanning revealed PM in 3 patients with normal chest X-rays.¹ The authors concluded that plain chest X-ray had a 10% false-negative rate. Similarly, other authors reported that plain chest X-ray alone was negative in 30% of SPM that was later diagnosed on CT scan.¹

Wu et al evaluated the necessity of a contrast fluoroscopic esophagogram among patients with evidence of PM on CT.¹ The study results suggest that contrast esophagograms are unnecessary if the CT is negative for oesophageal perforation or mediastinitis. Similar observations were reported by Neal et al.¹⁵ CT with on-table oral contrast has a high negative predictive value and is as sensitive as fluoroscopic oesophagography for diagnosing suspected oesophageal perforation. CT has greater sensitivity than fluoroscopic esophagography.¹ The fluoroscopic oesophagus does not provide any additional information that changes clinical management beyond what CT provides.¹ Other imaging modalities, such as ultrasound and magnetic resonance tomography, have been described in the literature; however, they do not appear to be superior to CT and, therefore, did not receive wide implementation.¹

Surgical interventions are not usually required for patients with SPM. Its use is reserved for patients with marked cardiorespiratory compromise or defined oesophageal or tracheal tear. If a tracheobronchial perforation is suspected due to blunt chest trauma, bronchoscopy is suggested. As observed by Neal et al, PM following blunt trauma in stable children is rarely associated with significant underlying injury to the tracheobronchial tree.¹⁵ Additionally, bronchoscopy can

aid in the localization and retrieval of foreign bodies and the evaluation of endobronchial lesions. Interestingly, Cunningham et al presented two paediatric cases with tracheal injury managed conservatively, suggesting that this may be a treatment option for some patients.¹⁶ The use of mediastinoscopy in alleviating life-threatening PM has been reported in a small number of cases. Chau et al. described percutaneous decompression of tension PM under fluoroscopic guidance using a drainage catheter and Heimlich valve in a 2-year-old girl with dermatomyositis and lung involvement.¹⁷ CT-guided placements may also be considered.¹⁸ Palmieri et al. described the technique of bedside supra jugular mediastinal decompression.¹⁹ The incision is placed in a suprasternal notch. After platysma muscle is dissected and deep cervical and pretracheal fascia are incised, a blunt dissection is made following the anterior surface of trachea, thus relieving tension PM.

SPM usually has benign course and resolves with supportive therapy only, but prolonged cases have also been reported (>2 months).¹ Minimizing physical activities that can precipitate SPM, such as weightlifting, and playing wind instruments, is recommended.¹ Patients are advised to quit scuba diving permanently.

Recurrence is extremely rare, with only four cases recorded in English literature.^{1,20} In such cases, it is reasonable to conduct additional diagnostic tests to detect any underlying lung or oesophageal pathologies. Some experts recommend performing lung function tests to detect any underlying pulmonary diseases that may have been missed.^{1,20} Medical conditions associated with the development of SPM, including asthma and recurrent vomiting from reflux disease, chemotherapy, cyclic vomiting, and bulimia, should be treated aggressively.

CONCLUSION

Three out of four patients presented to the ED had findings consistent with SPM without features of mediastinitis, hollow organ perforation or trauma. One patient had raised inflammatory markers and fever. According to current evidence, patients without features of mediastinitis or oesophageal perforation should not undergo further imaging and can be discharged after a short observation period. This approach was utilized in one of four cases. It is notable that the advice to proceed with further imaging was provided by surgeons as well as radiology consultants.

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