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CASE REPORT

# Spontaneous pneumomediastinum in two young women

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**Summary**
Spontaneous pneumomediastinum (SPM) is a rare benign condition which must be differentiated from secondary pneumomediastinum due to chest trauma, abscess formation or Boerhaave's syndrome. We present two young women with SPM due to chronic self-induced vomiting and starvation associated with psychosis and pregnancy-associated vomiting respectively. This report highlights the exclusionary diagnostic pathway, the principles of conservative management and the need for a tailored multidisciplinary approach to enhance patient recovery and prevent future recurrence.
**Keywords:** surgical emphysema, mediastinal emphysema

## Case 1

A 27-year-old woman presented with a three-month history of dysphagia for solids, inability to swallow, vomiting, heartburn and weight loss. There was no history of coughing, dyspnea, fever and trauma. She demonstrated persecutory delusions of being poisoned when eating and later admitted to self-induced vomiting due to this fear. On physical examination she was emaciated. The initial laboratory investigations showed hypokalaemia (2.5mmol/L), hypophosphataemia (0.43 mmol/L), hypomagnesaemia (0.54 mmol/L) and hypoalbuminaemia (30g/L) consistent with malnutrition and vomiting. Chest and lateral neck radiographs revealed subcutaneous emphysema, pneumomediastinum and prevertebral air (Figure 1). These findings, along with a small right pneumothorax were confirmed on computed tomography (CT) of the chest. Water-soluble swallow and upper gastrointestinal endoscopy (UGIE) were normal. The patient complained of inability to swallow, however swallowing assessment by speech therapy was normal. The diagnosis of spontaneous pneumomediastinum (SPM) was, therefore, made. The biochemical abnormalities were corrected with intravenous fluids and electrolytes and dietary supplements. She was referred to psychology and psychiatry in view of her fear of being given poisoned food and was diagnosed with a major depressive disorder with psychotic features. She did not meet diagnostic criteria for an eating disorder. On the eighth day of her admission her management was taken over by psychiatry who prescribed her an anti-psychotic, an anti-depressant and a benzodiazepine. She gained four kilograms over four weeks and was eating normally at follow-up.

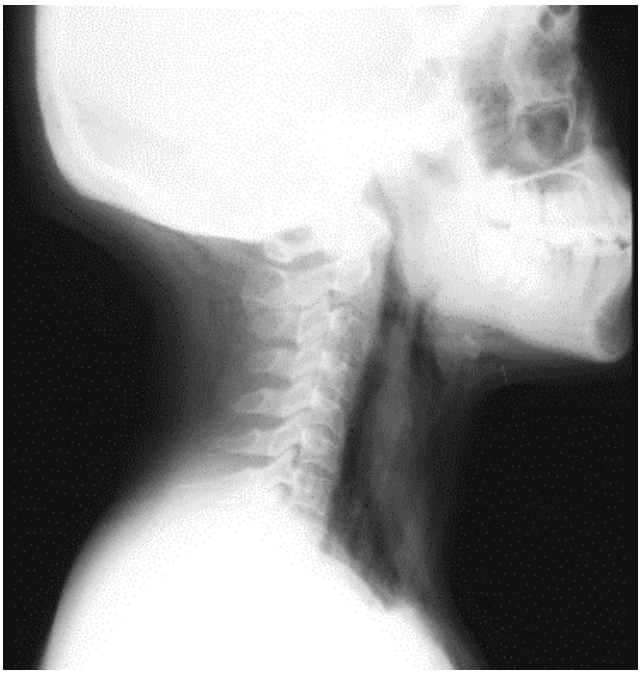

*Figure 1: Lateral neck X-ray showing prevertebral air*

## Case 2

An eight-week pregnant 24-year-old presented with a two-day history of persistent vomiting, odynophagia and dysphagia. There was no history of gastro-oesophageal reflux disease or corrosive ingestion. She had a slender physique, signs of dehydration and extensive surgical emphysema of her neck and chest. Her laboratory analysis showed hypokalaemia (2.8 mmol/L) and an acute kidney injury (urea: 15.8 mmol/L and creatinine: 220 umol/L). Chest and neck X-rays revealed a pneumomediastinum and prevertebral air. On CT scan, she had surgical emphysema, pneumomediastinum and pneumopericardium, (Figure 2). An inflamed oesophagus was seen on endoscopy. A water-soluble swallow showed no evidence of oesophageal leak.

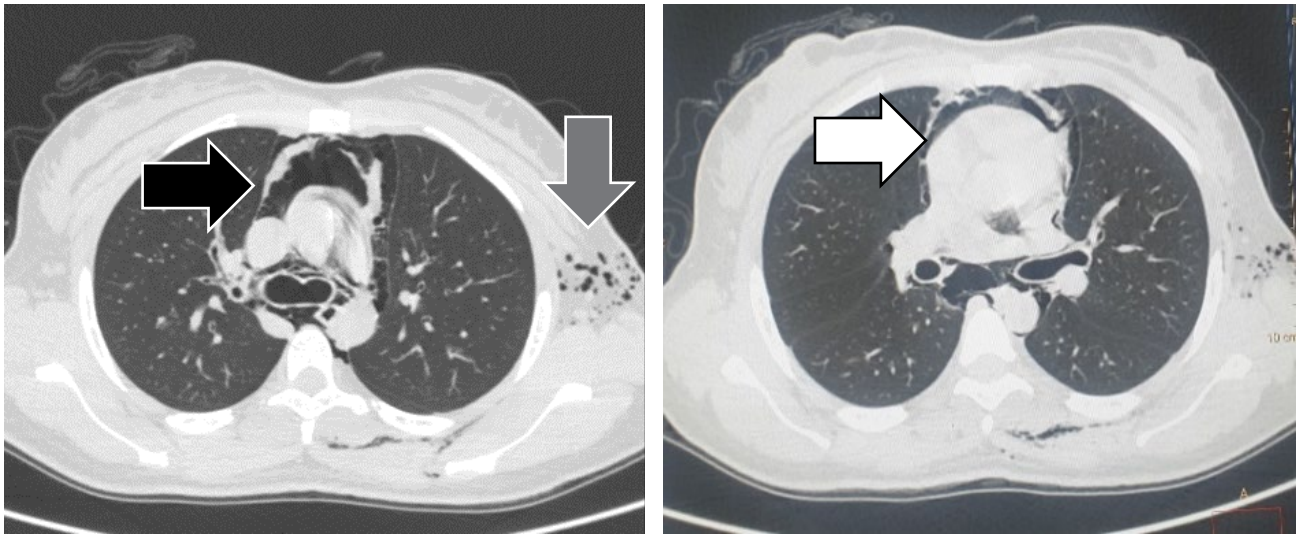

*Figure 2: CT scan axial views showing pneumomediastinum (black arrow), pneumopericardium (white arrow) and surgical emphysema (grey arrow)*

After exclusion of aerodigestive injury, she was diagnosed with SPM. Her laboratory abnormalities were corrected with intravenous fluids and potassium replacement. Initially, the patient was started on a broad-spectrum antibiotic whilst awaiting her water-soluble swallow to exclude oesophageal perforation. These were discontinued once this investigation was normal. Antiemetics were prescribed and an obstetric review organised for her pregnancy-associated vomiting in the event this would be an ongoing problem for her. Her symptoms and signs of SPM resolved by day two and she was discharged on day four. At two-week follow-up she had no complaints.

## Discussion

Over a two-month period, we admitted two young women with histories of vomiting, diagnosed with SPM. This is a rare, usually benign medical condition, characterised by free air in the mediastinum, without trauma, iatrogenic causes or underlying pulmonary pathology.[1] Initially described by Laennec in 1819, the syndrome was documented in postpartum women in 1939 by Louis Hamman.[2] SPM occurs after alveolar rupture following a period of increased intra-alveolar pressure. The interstitial air tracks along the bronchovascular sheaths toward the pulmonary hilum into the mediastinum and occasionally into the pericardium, the retropharyngeal and the retroperitoneal space.[3] This pathophysiology, described by Macklin in 1944, is known as the Macklin effect.[4] SPM may be associated with bronchial asthma, diabetic ketoacidosis, forceful straining during exercise, inhalation of drugs, childbirth, severe cough or vomiting, the Valsalva maneuver,[5] gastro-oesophageal reflux disease[6] and anorexia nervosa.[7]

Experimentally, caloric restriction has been shown to cause thinning of the alveolar walls[8] which may result in an air leak when there is an increase in the intra-alveolar pressure, as may occur with vomiting, as noted in our cases. The frequency of common signs and symptoms include: retrosternal chest pain (79%), subcutaneous emphysema (70%), neck pain (70%), dyspnea (58%), Hamman's sign—a crunching sound synchronous with the heartbeat—(52%), dysphagia (39%), back pain (8%), and epigastralgia (3%) were reported by Kaneki et al.[2,9] In this report, as in both our cases, patients were slender and presented with dysphagia and surgical emphysema, although 26 of their 33 patients were male.

Patients with SPM are haemodynamically stable and clinically well, indicating the benign nature of this condition. In 30% of cases, the diagnosis of SPM may be made on chest X-ray,[9] however, a CT scan should be done if there is diagnostic doubt or the suspicion of a perforated intra-thoracic viscus. A contrasted swallow may be valuable to rule out oesophageal perforation as may occur with Boerhaave's Syndrome. We investigated both our patients with CT scans as the diagnosis of SPM was not initially entertained since it is so rare in clinical practice. In view of the vomiting history, both patients had water-soluble swallows to rule out oesophageal perforation as the cause of their surgical emphysema and pneumomediastinum. The history and clinical examination excluded injury to the airway.

In a review of 34 patients with SPM by Ebina et al.[10] only two patients received antibiotics and none developed mediastinitis. We initiated antibiotics on our second patient whilst awaiting a water-soluble swallow to exclude oesophageal perforation, but these were discontinued when this was unremarkable. None of their or our patients developed a tension pneumomediastinum, delayed pneumothorax or airway compromise with conservative management, and in all, the SPM resolved spontaneously. Normal diet should be introduced once the patient feels comfortable, and the precipitating condition has been treated. Our patients average stay of 6 days was similar to the 4 to11 days reported in the Kaneki et al. series.[9]

## Conclusion

SPM is a rare clinical condition to be considered in a patient presenting with mediastinal emphysema without clinical signs of mediastinitis. Aerodigestive injury should be ruled out by imaging and/or endoscopy. Management of SPM is supportive and may warrant multidisciplinary input to treat the precipitating condition. In our cases, this was the psychiatrist and the obstetrician. SPM spontaneously resolves, and a good outcome can be expected.

### *Conflict of interest*

The authors declare no conflict of interest.


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### *Ethical approval*

Written informed consent was obtained from both patients in this case study. Ethics Committee approval was obtained from the University of KwaZulu-Natal Biomedical Research Ethics Committee BREC Ref No: BCA027/19.


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**REFERENCES**


1. Panack EA, Singer AJ, Sherman BW, Prescott A, Rutherford WF. Spontaneous pneumomediastinum: clinical and natural history. Ann Emerg Med. 1992;21(10):1222-7. https://doi.org/10.1016/s0196-0644(05)81750-0.
2. Hamman L. Spontaneous mediastinal emphysema. Bull Johns Hopkins Hosp. 1939;64:1-21.
3. Wintermark M, Wicky S, Schnyder P, Capasso P. Blunt traumatic pneumomediastinum: using CT to reveal the Macklin effect. AJR Am J Roentgenol. 1999; 172:129-30. https://doi.org/10.2214/ajr.172.1.9888752.
4. Macklin MT, Macklin CC. Malignant interstitial emphysema of the lungs and mediastinum as an important occult complication in many respiratory diseases and other conditions: interpretation of the clinical literature in the light of laboratory experiment. Medicine. 1944;23:281-358.
5. Murayama S, Gibo S. Spontaneous pneumomediastinum and Macklin effect: Overview and appearance on computed tomography. World J Radiol. 2014;6(11):850-4.https://doi.org/10.4329/wjr.v6.i11.850.
6. Huang HR, Wong KS, Lien R, Chiu CY. Spontaneous pneumomediastinum due to gastroesophageal reflux disease in an adolescent boy. Respirology. 2008;13:744-74. https://doi.org/10.1111/j.1440-1843.2008.01298.x.
7. Hochlehnert A, Löwe B, Bludau HB, et al. Spontaneous pneumomediastinum in anorexia nervosa: a case report and review of the literature on pneumomediastinum and pneumothorax. Eur Eat Disord Rev. 2010;18:107-15. https://doi.org/10.1002/erv.1002.
8. Massaro D, Massaro GD, Baras A, Hoffman EP, Clerch LB. Calorie-related rapid onset of alveolar loss, regeneration, and changes in mouse lung gene expression. Am J Physiol Lung Cell Mol Physiol. 2004;286:L896-906. https://doi.org/10.1152/ajplung.00333.2003.
9. Kaneki T, Kubo K, Kawashima, A et al. Spontaneous pneumomediastinum in 33 patients: yield of chest computed tomography for the diagnosis of the mild type. Respiration. 2000;67(4):408-11. https://doi.org/10.1159/000029539.
10. Ebina M, Inoue A, Takaba A, Ariyoshi K. Management of spontaneous pneumomediastinum: are hospitalization and prophylactic antibiotics needed? Am J Emerg Med. 2017;35(8):1150-53. https://doi.org/10.1016/j.ajem.2017.03.017.