Spontaneous pneumomedidastinum and pneumopericardium in a young, healthy adult with plans for international travel.

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### Abstract

A young man presented with sudden onset chest and neck pain, dyspnoea and dysphonia, following vigorous coughing. Examination identified surgical emphysema and subsequent CT scan revealed a spontaneous pneumomediastinum. The patient remained well despite the remarkable radiological findings. He was managed conservatively but his travel plans were interrupted.

## TITLE OF CASE

Spontaneous pneumomediastinum and pneumopericardium in a young, healthy adult with plans for international travel.

### KEY CLINICAL MESSAGE

Although rare, pneumomediastinum and pneumopericardium should be considered in patients presenting with sudden onset chest discomfort after coughing.

## **AUTHOR CONTRIBUTION**

 $1^{st}$  Author Dr J Winterton – writing up of case report including literature review and liaising with patient  $2^{nd}$  Author Dr S Biart – revision and edition of case report

# **SUMMARY**

A well 23 year old male presented complaining of sudden onset chest and neck pain, dyspnoea and dysphonia, following an episode of vigorous coughing. Clinical examination revealed a mild tachycardia and evidence of surgical emphysema in the left supraclavicular fossa. A chest x-ray confirmed surgical emphysema in the left side of the neck and raised concern of pneumomediastinum. The subsequent CT scan identified both pneumomediastinum and more surprisingly, pneumopericardium. No pneumothorax was identified. This spontaneous event was treated conservatively with analgesia and observation. The patient remained remarkably well and was discharged after 48 hours with safety net advice and a plan for appropriate follow up. His travel plans were complicated as the treating team advised against imminent air travel, although specific guidelines for this condition are lacking.

## CASE PRESENTATION

A 23 year old male presented to the emergency department with sudden onset pleuritic chest pain and associated left sided neck pain. He complained of dyspnoea, whilst also feeling his voice had gained a higher pitch. The onset of these symptoms came following a vigorous episode of coughing, in conjunction with three days of coryzal symptoms. He had no past medical history, took no regular medications and was not

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known to have any allergies. He did not smoke or use illicit substances for recreational use, drank minimal alcohol and was physically active. There was no family history of note. On examination the gentleman was tall with a medium build. On admission, he was in some discomfort but was haemodynamically stable and had no respiratory compromise. There were no signs of tamponade and his heart sounds were audible. He had surgical emphysema palpable in the left supraclavicular fossa, but had a central trachea and clear lung fields with normal resonance.

#### INVESTIGATIONS

The patient's inflammatory markers were mildly raised. An ECG showed a sinus tachycardia which resolved. An initial radiograph identified surgical emphysema extending in to the neck from the left lung apex with associated pneumomediastinum (Figure 1). This warranted an urgent CT with contrast oesophagealogram in a hope to localise the source of the free air. This confirmed the presence of surgical emphysema and pneumomediastinum (Figure 2), but also revealed pneumopericardium (Figure 3). No source of the free air was identified.

### DIFFERENTIAL DIAGNOSIS

With the presence of sudden onset pain, dyspnoea and surgical emphysema, a pneumothorax was the primary differential on admission. The spontaneous, abrupt onset would raise suspicion also for pulmonary embolism, however the patient had no risk factors, haemoptysis or objective hypoxia. The radiological suggestion of pneumomediastinum was surprising, but perhaps explained the dysphonia reported. Once identified, a source for the pneumomediastinum and pneumopericardium was sought; CT scan ruled out Boerhave oesophageal rupture, soft tissue infection or lung pathology. Other cases of isolated pneumopericardium published have been related to perforated gastric ulcers, but this young man had no symptoms or signs suggestive of this. [11REFERENCESGossage AA, Robertson PW, Stephenson SF Spontaneous pneumopericardium. Thorax 1976;31:460-465] A life-threatening sequelae of free air in the pericardial space is tension pneumopericardium resulting in tamponade, however the patient remained haemodynamically stable with no clinical evidence of this.

## TREATMENT

The patient was admitted and treated with adequate analgesia. Given he had a productive cough and raised inflammatory markers he was started on oral antibiotics for a lower respiratory tract infection. The case was discussed with cardiothoracic specialists. Following review of the CT scan, it was suggested that the most probable cause of the presentation was a ruptured pulmonary bleb resulting from forceful coughing. Although usually attributed to the formation of spontaneous pneumothorax, this was thought to be the origin of the pneumomediastinum and pneumopericardium. The parent team was advised to treat conservatively and monitor for a further 24 hours to ensure the surgical emphysema was not increasing in size - which would suggest ongoing air leak.

The patient was counselled regarding the life-time contraindication to scuba diving and the need to avoid flying until resolution of free air was confirmed at follow up. There is a lack of guideline or evidence for management of pneumomediastinum and pneumopericardium therefore guidelines on pneumothorax were adapted – the risks of tension and tamponade from rapid pressure change theoretically remains the same. The patient was observed for a period, and discharged with safety net advice.

### OUTCOME AND FOLLOW-UP

The admission was a frustrating one for the patient, who despite feeling well advised to remain in for observation initially. After discharge, follow up was arranged with respiratory physicians with repeat CXR. Although a plain chest X-ray can normally be used to see resolution of a pneumothorax or even pneumomediastinum, this would not adequately ensure resolution of pneumopericardium and further CT imaging would be necessary. The patient suffered from ongoing discomfort and shortness of breath initially after discharge but this soon settled.

Unfortunately, the patient had extensive travel plans set for the weeks following discharge. These had to be postponed on medical advice. Careful communication of the potential sequalae of air travel was required. In addition advice on a life-long avoidance of scuba diving was a blow to the patient.

Follow up of the patient to gain further patient perspective involved several telephone conversations between the author and the case subject. Interestingly, he was happy to speak to the author, however was not keen on attending for follow up CT scan or consultation with the respiratory physicians. From the patient's perspective he had been primarily concerned with follow up identify incomplete resolution of the pneumopericardium and was anxious that the medical team would inform him air travel was contraindicated. This was significant for him as a young, healthy adult who had planned and paid for a period of international travel involving multiple flights. Encouragement to attend follow up was made, however the patient was adamant he did not want to attend and felt that because his symptoms had resolved, he would rather take the risk. His autonomy was respected.

## **DISCUSSION**

Spontaneous pneumomediastinum (SPM) is defined as free air within the mediastinum, without an identifiable cause. The condition may be referred to as Hamman syndrome, following the first reported case in 1939.[11Hamman L. Spontaneous mediastinal emphysema. Bull Johns Hopkins Hosp. 1939;64:1–21] To be classed as spontaneous, clear extrinsic causes must be ruled out; most predominantly recognised are oesophageal rupture (Booerhave's), infection with gas producing organisms and trauma. The dissection of alveolar air through the interstitium of the lung, along the bronchovascular sheath to the hilum is known as the Macklin effect. [2] [22Macklin 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. Ruptured alveoli can result from increased broncho-alveolar pressure. This phenomenon is attributed to a range of causes, including the hyperventilation of asthma or those in diabetic ketoacidosis, performing the valsalveur manoeuvre, substance inhalation or as in this case, vigorous coughing. [33Newcomb AE, Clarke CP. Spontaneous pneumomediastinum: a benign curiosity or a significant problem? Chest. 2005;128:3298– 3302][44Weathers LS, Brooks WG, DeClue TJ. Spontaneous pneumomediastinum in a patient with diabetic ketoacidosis: a potentially hidden complication. South Med J. 1995;88:483–484.][55Beauchamps G. Spontaneous pneumothorax and pneumomediastinum. In: Pearson FG, Deslauriers J, Ginsberg RJ, Hiebert CA, McKneally MF, et al., editors. Thoracic surgery. New York: Churchill Livingstone; 1995. pp. 1037-1054.][66Jougon JB, Ballester M, Delcambre F, Mac Bride T, Dromer CE, Velly JF. Assessment of spontaneous pneumomediastinum: experience with 12 patients. Ann Thorac Surg. 2003;75:1711–1714]

SPM affects predominantly males aged 17 to 25 and has an estimated incidence of 1 in 30,000 hospital admissions. [77Kaur H, Singh G, Aggarwal S, Singh A. Spontaneous pneumomediastinum with pneumopericardium, surgical emphysema, pneumothorax, and epidural pneumotosis: A rare association. Journal of Natural Science, Biology and Medicine. 2014;5(1):201.] We recognise this figure is unlikely to reflect true incidence given the rarity of the disease and that its transient nature predisposes to poor levels of observation. As in this case, patients with SPM will most commonly present complaining of chest pain and shortness of breath with associated subcutaneous emphysema of the neck. [88Macia I, Moya J, Ramos R et al. Spontaneous pneumomediastinum: 41 cases. Eur J Cardiothorac Surg2007;31:1110–14] Although no official guideline for diagnosis exists, CT scan including an oral contrast study to rule out oesophageal rupture is suggested as the favourable method of investigation, given 70% of SPM is missed on plain chest radiograph. [4]

Cases of spontaneous pneumopericardium in conjunction with pneumomediastinum are extremely rare. A medline search with the terms 'spontaneous pneumomediastinum' AND 'pneumopericardium' returned 40 results. Of these, 11 were truly case reports of adults with spontaneous pneumomediastinum with associated spontaneous pneumopericardium. We struggled to find other cases which described such a mild symptom profile coupled with such significant radiological findings. This was of interest as it contributed to our feeling that this pathology could be misdiagnosed or missed altogether with relative ease in a young, healthy population. Pneumopericardium, although requiring higher pressures to develop than SPM, is again most likely

caused by air dissecting through the lung interstitium and medially along the venous sheaths into the pericardial space. [99Lee Y, Jin S, Jang S, Jang Y, Lee E, Kim Y et al. A Case of Spontaneous Pneumomediastinum and Pneumopericardium in a Young Adult. The Korean Journal of Internal Medicine. 2001;16(3):205-209.] There is no clinical guideline for the management of SPM or pneumopericardium. Both conditions can follow a benign course, suggested treatment being with conservative measures such as analgesia and observation in addition to appropriate investigation of the cause. [10]

Pneumopericardium may be complicated by tension. Although mentioned only in cases of traumatic pneumopericardium, we recognise that the spontaneous form of this diagnosis still carries this risk.[1010Capizzi PJ, Martin M, Bannon MP. Tension pneumopericardium following blunt injury. J Trauma 1995;39:775–80] This is a life-threatening condition leading to cardiac tamponade and risk of haemodynamic instability and arrest. Tension pneumopericardium should therefore be excluded once diagnosis of free air in the pericardial space is made. Hemodynamic instability clearly raises high suspicion. ECG has been suggested as beneficial in early recognition of this diagnosis.[1111Minns T, Raj R, Clark K A pain in the neck Case Reports 2011;2011:bcr0920114840.] Non-specific ST and T wave changes and decreased amplitude on ECG have been recognised in at least two cases of tension pneumopericardium.[1212Lucarelli K, Troisi F, Langialonga T. Pneumopericardium. Eur Heart J 2010;31:1953][1313Konijn AJ, Egbers PH, Kuiper MA. Pneumopericardium should be considered with electrocardiogram changes after blunt chest trauma: a case report. J Med Case Reports 2008;2:100

#### **FIGURES**

**Figure 1**: Admission chest x-ray demonstrating pneumomediastinum and surgical emphysema in the left supraclavicular fossa.

Figure 2: CT scan slice demonstrating pneumomediastinum.

Figure 3: CT scan slice demonstrating pneumopericardium.]

## LEARNING POINTS/TAKE HOME MESSAGES

- Pneumomediastinum and pneumopericardium should be a consideration in adult patients presenting with sudden onset chest pain the transient and mild nature of symptoms in young people do no correlate well with the severity of pathological findings on radiological imaging and may contribute to under-diagnosis.
- The management of spontaneous pneumomediastinum and pneumopericardium is largely conservative, but clinicians should be vigilant for life-threatening sequelae of tension pneumopericardium.
- The impact of this diagnosis on the patient was profound and restrictive. Clinicians should be clear in communicating risk, in this case adapted from pneumothorax guidelines. The patient's perspective and priorities may be different than expected.





