

A review of non-obstetric spontaneous pneumomediastinum and subcutaneous emphysema

M. BRAND, M.B. CH.B., F.C.S. (S.A.), M.R.C.S., M.MED. (SURG.)

B. BIZOS, M.B. B.CH., F.C.S. (S.A.), F.R.C.S. (ENG.). M.MED. (SURG.)

Department of Surgery, University of the Witwatersrand, Johannesburg

L. BURNELL, M.B. B.CH.

Department of Ear, Nose and Throat Surgery, University of the Witwatersrand

A man in his early twenties was admitted from the streets. He was hallucinating and incoherent. His vital signs were normal and there was no sign of injury. There was subcutaneous emphysema over the neck and anterior chest. The breath sounds were good and the heart sounds were audible. Chest X-ray showed extensive subcutaneous emphysema extending into the neck, but no pneumomediastinum or pneumothorax. A Hexabrix swallow demonstrated no leak. A computed tomography (CT) scan showed extensive surgical emphysema but no underlying pathology. Over the following 48 hours he was treated with a 40% oxygen mask. He remained stable and the surgical emphysema settled completely. Urine testing for cannabis and cocaine were negative. He was certified and admitted to a closed psychiatric ward. No further surgical problems occurred during the following month.

Review

Louis Hamman originally reported the syndrome that bears his name in 1939,¹ describing it in women in labour ('postpartum pneumomediastinum'). More recently, the syndrome has been loosely applied to all cases of spontaneous pneumomediastinum and subcutaneous emphysema. This review includes an overview of 561 cases, nearly 10 times more than in any other study.

Spontaneous pneumomediastinum (SPM) is an uncommon and usually benign entity, characterised by the presence of free air in the mediastinum, in the absence of traumatic or iatrogenic causes or preceding pulmonary pathology such as emphysema, chronic bronchitis or lung cancer.² Macklin³ elucidated the pathophysiology of this condition based on animal laboratory studies in 1944; this was revised by Maunder *et al.*⁴ SPM produces a dramatic increase of endopulmonary pressure⁵ that results in rupture of alveolar septa, which causes interstitial air to extend along peribronchial and perivascular spaces into the mediastinum.^{3,4} Frequently, air extends to the neck along the cervical fascia, developing into subcutaneous emphysema.⁶ Valsalva manoeuvres have been implicated as the cause of SPM.⁷⁻⁹ If prolonged, they result in acute, intermittent lower airway obstruction and increased intra-alveolar pressure resulting in rupture of alveoli.¹⁰

'Hamman's crunch' (Hamman's sign) is a precordial crunching sound, heard best when the patient is in the left lateral decubitus position.¹ It is, however, not pathognomonic, and can occur with bullous emphysema, pneumothorax and dilatation of the distal oesophagus.⁸

Pneumomediastinum is seen on chest X-ray. On P-A projections there are radiolucent streaks in the mediastinum that can surround the pericardium and reveal the thin dense line of the anterior mediastinal pleura. The lateral view often demonstrates retrosternal free air, with clear definition of mediastinal structures such as the aorta.¹¹

Before spontaneous subcutaneous emphysema and pneumomediastinum can be attributed to 'Hamman's syndrome', potentially lethal causes should be considered and excluded. Some of these conditions are:

- oesophageal: rupture, spasm, oesophagitis
- cardiac: ischaemia, pericarditis
- pulmonary: embolism, pneumothorax, pneumonia
- musculoskeletal: costochondritis, inflammatory joint disorders.

Most can be excluded with a good history, thorough clinical examination and targeted special investigations. Munsell⁸ maintained that the presence of subcutaneous emphysema without apparent cause, together with pneumomediastinum on chest X-ray, was sufficient to diagnose SPM.

Treatment includes the administration of 95% oxygen to relieve the dyspnoea and to facilitate the reabsorption of nitrogen.¹⁰ The oxygen decreases the partial pressure of nitrogen in the blood, which then promotes the diffusion of nitrogen from the interstitium back into the blood. This increases the resorption of the mediastinal air.¹²

We performed a Pubmed search to identify and review all cases of non-obstetric SPM. The search was confined to English-language journals, and the only initial limit was the exclusion of paediatric cases. Key words were Hamman's syndrome, spontaneous pneumomediastinum and spontaneous subcutaneous emphysema. This resulted in 295 references. All papers with obstetric cases were excluded. We also excluded papers in which there was a predisposing condition, or possible aetiological agent, since these were not spontaneous, as well as papers without case studies. This resulted in 57 articles^{2,5-7,9-11,13-61} for analysis. We were able to acquire all the original articles.

A total of 561 non-obstetric patients were included in our analysis; the majority of papers were reports of a single case. The mean patient age was 23 years and 70.2% were male. Most cases were related to activities that resulted in a prolonged Valsalva manoeuvre such as coughing, strenuous sport, or the use of inhaled drugs

such as cocaine and marijuana (10%). Psychiatric disorders were reported in 6% of patients, but the association is not clear; it may in fact be the consequence of an unrecognised Valsalva episode.

The three most common symptoms were chest pain (61.3%), dyspnoea (38.3%) and dysphagia (14.5%). Clinical examination revealed subcutaneous emphysema (56.1%) and Hamman's crunch (17.1%). Of note is that subcutaneous emphysema only occurred in half the patients, and that Hamman's sign was positive in less than 1 in 5 patients.

As far as radiological investigations were concerned, chest X-ray was diagnostic in 76.4%. Chest CT scans were positive in all of the 172 scans performed (i.e. 100%); not a single oesophagogram showed any contrast leak (211 performed); and the bronchoscopies performed (108) were uniformly negative. The inference is that, given a suggestive history and adequate CT studies, invasive procedures are not required.

Management was successful using high concentrations of oxygen in all cases. Only 3.1% of patients required an intercostal drain for pneumothorax. The mean hospital stay was 3.5 days. There was only 1 death, in a patient who developed a stroke during his admission. There were 7 (1.3%) recurrences: 3 in scuba divers, and 1 in a skydiver. The remaining 3 recurrences appear to have been genuinely spontaneous as there was no repetitive activity.

Conclusion

Non-obstetric SPM is a benign entity that should be diagnosed when the history suggests a prolonged Valsalva manoeuvre, with cervical or thoracic subcutaneous emphysema. A chest X-ray should confirm the pneumomediastinum and subcutaneous emphysema, but chest CT is the standard for diagnosis. Invasive investigations are usually not required. Management is conservative, with oxygen; all cases resolve spontaneously. Intercostal drain insertion for a pneumothorax is seldom required. Recurrence is rare, but individuals involved in a professional sport that induces repetitive prolonged Valsalva manoeuvres should be warned that they could possibly experience a recurrence.

Acknowledgements

Mr J Krabenius of the World Gastroenterological Organization Library in Paris was of enormous help in locating many of the obscure references listed below.

In all, 61 papers were analysed for this review. The first 12 and most important references are listed in the printed version of the SAJS, while the full number have been included in the online version of the August 2011 SAJS, which is available at the open-access website www.sajs.org.za.

REFERENCES

- Hamman L. Spontaneous mediastinal emphysema. *Bull Johns Hopkins Hosp* 1939;64:1-21.
- Panacek EA, Singer AJ, Sherman BW, Prescott A, Rutherford WF. Spontaneous pneumomediastinum: clinical and natural history. *Ann Emerg Med* 1992;21(10):1222-1227.
- 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.
- Maunder RJ, Pierson DJ, Hudson LD. Subcutaneous and mediastinal emphysema. Pathophysiology, diagnosis and management. *Arch InternMed* 1984;144:1447-1453.
- Jougon 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(6):1711-1714.
- Mondello B, Pavia R, Ruggeri P, Barone M, Barresi P, Monaco M. Spontaneous pneumomediastinum: experience in 18 adult patients. *Lung* 2007;185(1):9-14.
- Steffey WR, Cohn AM. Spontaneous subcutaneous emphysema of the head, neck and mediastinum. *Arch Otolaryngol* 1974;100:32-35.
- Munsell WP. Pneumomediastinum. *JAMA* 1967;202:689-693.
- Mattox KL. Pneumomediastinum in heroin and marijuana users. *JACEP* 1976;5:26-28.
- Hunter JG, Loy HC, Markovitz L, Kim US. Spontaneous pneumomediastinum following inhalation of alkaloidal cocaine and emesis: case report and review. *Mt Sinai J Med* 1986;53(6):491-493.
- Halperin AK, Deichmann RE. Spontaneous pneumomediastinum: a report of 10 cases and review of the literature. *NC Med J* 1985;46(1):21-23.
- Fine J, Frelings S, Starr A. Experimental observations on the effects of 95% oxygen on the absorption of air from the body tissues. *J Thoracic Surgery* 1935;4:635.
- Aldridge SM, Glover SC, Johnson C. Spontaneous pneumomediastinum in two stowaways. *Br Med J (Clin Res Ed)* 1986;293(6541):243.
- Al-Mufarrej F, Badar J, Gharagozloo F, Tempesta B, Strother E, Margolis M. Spontaneous pneumomediastinum: diagnostic and therapeutic interventions. *J Cardiothorac Surg* 2008;3:59.
- Ba-Salamah A, Schima W, Umek W, Herold CJ. Spontaneous pneumomediastinum. *Eur Radiol* 1999;9(4):724-727.
- Bratton SL, O'Rourke PP. Spontaneous pneumomediastinum. *J Emerg Med* 1993;11(5):525-529.
- Cheung HY, Law S, Wong KH, Kwok KF, Wong J. Spontaneous pneumomediastinum in a scuba diver. *Hong Kong Med J* 2006;12(2):152-153.
- Dickie HA. Spontaneous mediastinal emphysema and spontaneous pneumothorax; a report of 20 cases. *Ann Intern Med* 1948;28(3):618-629.
- Fajardo LL. Association of spontaneous pneumomediastinum with substance abuse. *West J Med* 1990;152(3):301-304.
- Fergusson RJ, Shaw TR, Turnbull CM. Spontaneous pneumomediastinum: a complication of anorexia nervosa? *Postgrad Med J* 1985;61(719):815-817.
- Forshaw MJ, Khan AZ, Strauss DC, Botha AJ, Mason RC. Vomiting-induced pneumomediastinum and subcutaneous emphysema does not always indicate Boerhaave's syndrome: report of six cases. *Surg Today* 2007;37(10):888-892.
- Freixinet J, Garcia F, Rodriguez PM, Santana NB, Quintero CO, Hussein M. Spontaneous pneumomediastinum long-term follow-up. *Respir Med* 2005;99(9):1160-1163.
- Fugo JR, Reade CC, Kypson AP. Spontaneous pneumomediastinum. *Curr Surg* 2006;63(5):351-353.
- Grant T. Primary (spontaneous) pneumomediastinum presenting as abdominal pain after swimming. *Ann Emerg Med* 1988;17(12):1368-1369.
- Grossman A, Romem A, Azaria B, Goldstein L, Barenboim E. Pneumomediastinum in student aviators: 10 cases with return to flying duty. *Aviat Space Environ Med* 2005;76(1):63-65.
- Gunluoglu MZ, Cansever L, Demir A, et al. Diagnosis and treatment of spontaneous pneumomediastinum. *Thorac Cardiovasc Surg* 2009;57(4):229-231.
- Hatzitolios A, Ntaios G. Spontaneous pneumomediastinum may be associated with both anorexia nervosa and obesity. *Lung* 2007;185(6):373.
- Herlan DB, Landreneau RJ, Ferson PF. Massive spontaneous subcutaneous emphysema. Acute management with infraclavicular "blow holes". *Chest* 1992;102(2):503-505.
- Iyer VN, Joshi AY, Ryu JH. Spontaneous pneumomediastinum: analysis of 62 consecutive adult patients. *Mayo Clin Proc* 2009;84(5):417-421.
- James M, Miguel M, Fancher T. Spontaneous pneumomediastinum. *J Hosp Med* 2007;2(4):283-284.
- Adwers JR, Hodgson PE, Lynch R. Spontaneous pneumomediastinum. *J Trauma* 1974;14(5):414-418.
- Kaneki T, Kubo K, Kawashima A, Koizumi T, Sekiguchi M, Sone S. Spontaneous pneumomediastinum in 33 patients: yield of chest computed tomography for the diagnosis of the mild type. *Respiration* 2000;67(4):408-411.
- Kosaka T, Haraguchi M, Tsuneoka N, Furui J. Spontaneous pneumomediastinum as a result of SCUBA diving. *Eur J Emerg Med* 2007;14(2):118-119.
- Lin LY, Kwok CF, Tang KT, Ho LT, Lin HD. Diffuse soft tissue emphysema in anorexia nervosa: a case report. *Int J Eat Disord* 2005;38(3):277-280.
- Majer S, Graber P. Postpartum pneumomediastinum (Hamman's syndrome). *CMAJ* 2007;177(1):32.
- Mazur S, Hitchcock T. Spontaneous pneumomediastinum, pneumothorax and ecstasy abuse. *Emerg Med (Fremantle)* 2001;13(1):121-123.
- Nichols AW. Spontaneous pneumomediastinum in a collegiate volleyball player. *Clin J Sport Med* 1999;9(2):97-99.
- Pangtey GS, Das CJ, Javan N. Airlessness in airspace. Simultaneous occurrence of spontaneous pneumothorax with pneumomediastinum and pneumorrhachis: report of a case. *Surg Today* 2008;38(1):49-51.
- Parker GS, Mosborg DA, Foley RW, Stiernberg CM. Spontaneous cervical and mediastinal emphysema. *Laryngoscope* 1990;100(9):938-940.
- Badaoui R, El Kettani C, Filki M, Ouendo M, Canova-Bartoli P, Ossart M. Spontaneous cervical and mediastinal air emphysema after ecstasy abuse. *Anesth Analg* 2002;95(4):1123.
- Chu CM, Leung YY, Hui JY, et al. Spontaneous pneumomediastinum in patients with severe acute respiratory syndrome. *Eur Respir J* 2004;23(6):802-804.
- Ring A, Liebert T, Stern J. Pneumopericardium after hyperemesis: Possible result of the Macklin effect. *Chirurg* 2010;81(6):568-571.
- Esayag Y, Furer V, Izbicki G. Spontaneous pneumomediastinum: is a chest X-ray enough? A single-center case series. *Isr Med Assoc J* 2008;10(8-9):575-578.
- Townes DA. Spontaneous pneumomediastinum in a marathon runner. *Br J Sports Med* 2006;40(10):878-879.
- Weissberg D, Weissberg D. Spontaneous mediastinal emphysema. *Eur J Cardiothorac Surg* 2004;26(5):885-888.

46. Tytherleigh MG, Connolly AA, Handa JL. Spontaneous pneumomediastinum. *J Accid Emerg Med* 1997;14(5):333-334.
47. Takada K, Matsumoto S, Hiramatsu T, et al. Management of spontaneous pneumomediastinum based on clinical experience of 25 cases. *Respir Med* 2008;102(9):1329-1334.
48. Newcomb AE, Clarke CP. Spontaneous pneumomediastinum: a benign curiosity or a significant problem? *Chest* 2005;128(5):3298-3302.
49. Mortelmans LJ, Bogaerts PJ, Hellemans S, Volders W, Van Rossom P. Spontaneous pneumomediastinum and myocarditis following Ecstasy use: a case report. *Eur J Emerg Med* 2005;12(1):36-38.
50. Teixeira Moreira Almeida Mdo S, Dias LT, Fernandes SJ, Almeida JV. Spontaneous pneumomediastinum and subcutaneous emphysema in systemic sclerosis. *Rheumatol Int* 2007;27(7):675-677.
51. Caceres M, Ali SZ, Braud R, Weiman D, Garrett HE Jr. Spontaneous pneumomediastinum: a comparative study and review of the literature. *Ann Thorac Surg* 2008;86(3):962-966.
52. Koullias GJ, Korkolis DP, Wang XJ, Hammond GL. Current assessment and management of spontaneous pneumomediastinum: experience in 24 adult patients. *Eur J Cardiothorac Surg* 2004;25(5):852-855.
53. Macia I, Moya J, Ramos R, et al. Spontaneous pneumomediastinum: 41 cases. *Eur J Cardiothorac Surg* 2007;31(6):1110-1114.
54. Abolnik I, Lossos IS, Breuer R. Spontaneous pneumomediastinum. A report of 25 cases. *Chest* 1991;100(1):93-95.
55. Morgan J, Sadler MA, Yeghiayan P. Spontaneous pneumomediastinum in a patient with recent air travel. *Emerg Radiol* 2007;14(6):457-459.
56. Nicol E, Davies G, Jayakumar P, Green ND. Pneumopericardium and pneumomediastinum in a passenger on a commercial flight. *Aviat Space Environ Med* 2007;78(4):435-439.
57. James M, Miguel M, Fancher T. Spontaneous pneumomediastinum. *J Hosp Med* 2007;2(4):283-284.
58. Maravelli AJ, Skiendziewski JJ, Snover W. Pneumomediastinum acquired by glass blowing. *J Emerg Med* 2000;19(2):145-147.
59. Frenkel MA, Lyons LL. Spontaneous pneumomediastinum. An unusual cause of a sore throat. *Postgrad Med* 1991;89(1):257-259.
60. Zaia BE, Wheeler S. Pneumomediastinum after inhalation of helium gas from party balloons. *J Emerg Med* 2010;38(2):155-158.
61. Kikuchi N, Ishii Y, Satoh H, Ohtsuka M, Hizawa N, Ohta Y. Spontaneous pneumomediastinum after air travel. *Am J Emerg Med* 2008;26(1):116.e1-2. Qureshi SA, Tilyard A. Unusual presentation of spontaneous pneumomediastinum: a case report. *Cases J* 2008;1(1):349.