

CASE REPORT

Simultaneous bilateral spontaneous hydropneumothorax: a rare presentation of bilateral malignant pleural mesothelioma

Hossam E Fayed, Victoria K Woodcock, Jamal Grayez

Department of Medicine,
Oxford University Hospitals,
Banbury, UK

Correspondence to
Dr Hossam E Fayed,
hssmfayed@yahoo.com

SUMMARY

This is a case of a 69-year-old man with a history of asbestos exposure who presented with acute shortness of breath. His chest x-ray showed bilateral hydropneumothorax. Further investigations including CT chest and video-assisted thoracoscopic surgery revealed bilateral pleural thickening and histology confirmed epithelioid mesothelioma. This case highlights the need for clinicians to be aware of atypical presentations of malignant pleural mesothelioma as well as the importance of considering underlying secondary causes such as malignancy in the older patient presenting with spontaneous pneumo/hydropneumothorax.

BACKGROUND

Malignant pleural mesothelioma (MPM) is a rare malignancy with a poor prognosis. It typically presents in patients with a history of asbestos exposure in the fifth to seventh decades of life with symptoms of dyspnoea, non-pleuritic chest pain or both. The most common radiographical presentation is unilateral pleural effusion with or without ipsilateral pleural thickening or mass.

CASE PRESENTATION

A previously fit and healthy 69-year-old man presented with a 6-day history of shortness of breath on exertion and fleeting non-pleuritic pain in his chest. He denied any symptoms of cough, haemoptysis, fever or weight loss, and had no orthopnoea or ankle swelling. He had a history of long-haul air travel 3 weeks previously, but no symptoms of calf pain or swelling. He was a lifelong non-smoker and was an electrician by trade, with a history of asbestos exposure between the age of 15 and 30 years.

On examination he was neither tachycardic nor tachypnoeic and had oxygen saturations of 94% at room air. Chest auscultation revealed reduced breath sounds bilaterally.

INVESTIGATIONS

A chest radiograph revealed bilateral hydropneumothorax (figures 1 and 2). A CT chest with pulmonary angiography was performed to look for potential secondary causes of pneumothorax.

The CT chest showed large bilateral hydropneumothorax with significant collapse of the underlying lungs (figure 3). There was a very small amount of mediastinal free gas and a 10 mm superior mediastinal, 12 mm right hilar and 15 mm

subcarinal node. There was bilateral thickening more marked on the left.

TREATMENT

The patient had bilateral chest drains inserted. Diagnostic cytology of the pleural fluid showed moderate numbers of reactive mesothelial cells, many polymorphs, some lymphocytes and red blood cells but no malignant cells. The patient underwent video-assisted thoracoscopic surgery (VATS) which showed pleural nodules and fibrinous deposits, more on the left than on the right. Talc pleurodesis was performed in the same setting. Pleural biopsies revealed epithelioid mesothelioma.

OUTCOME AND FOLLOW-UP

The patient was referred to oncology and underwent four cycles of cisplatin and pemetrexed chemotherapy, for which he responded well both clinically and radiologically on follow-up imaging. However, 11 months after his diagnosis, further imaging showed left-sided circumferential pleural thickening and progression of the disease. (figure 4). Unfortunately, the patient died 4 months later.

DISCUSSION

Radiologically, MPM typically presents with pleural effusion and pleural thickening. Although uncommon, spontaneous pneumothorax is a recognised

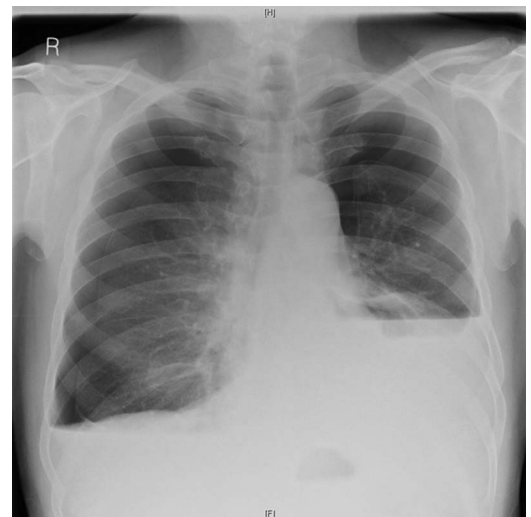


Figure 1 Anteroposterior view of chest radiograph showing bilateral hydro pneumothorax.

To cite: Fayed HE, Woodcock VK, Grayez J. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-009350



Figure 2 Lateral view of chest radiograph showing bilateral hydropneumothorax.

presentation.¹ Hydropneumothorax secondary to MPM is also uncommon with descriptions of this phenomenon limited to a handful of case reports.^{2–5} The mechanism underlying such presentations is uncertain but may be related to rupture of necrotic tumour nodules.

Bilateral presentations of pneumothorax and hydropneumothorax are even more unusual.

On review of literature we were able to find only one case report describing bilateral spontaneous pneumothorax as the primary presentation of malignant pleural mesothelioma.⁶ We were unable to find any cases describing bilateral hydropneumothorax in mesothelioma, and indeed there appear to be only two documented cases of bilateral hydropneumothorax because of other causes. Steeghs *et al*⁷ reported a case in a patient with pulmonary rheumatoid nodules during treatment with methotrexate, while Daoud⁸ reported a similar case in pulmonary sarcoidosis.

Given that the number of mesothelioma cases in the UK has risen in the past 50 years, with deaths predicted to peak between 2011 and 2015,⁹ recognition of atypical presentation is increasingly important to facilitate timely diagnosis and appropriate management. In a recent case series Saleh *et al*² suggest performing CT scans of the chest in all patients over the age of



Figure 3 CT chest showing bilateral hydropneumothorax and pleural thickening.

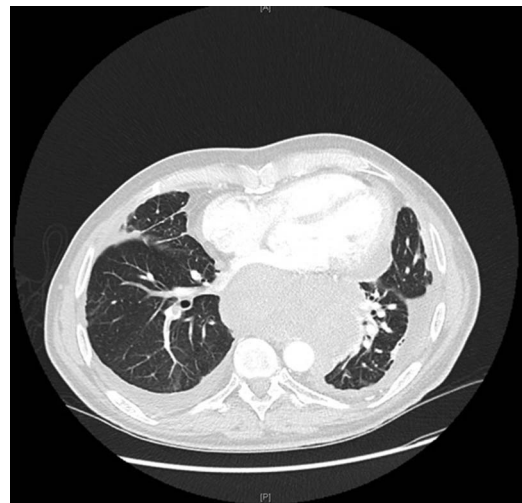


Figure 4 CT chest showing bilateral progression of the mesothelioma.

50 presenting with spontaneous pneumothorax so as to rule out an underlying malignant cause or visualise big bullae. Although there are currently no recommendations to this effect in the UK BTS¹⁰ or American College of Chest Physicians¹¹ guidelines on management of spontaneous pneumothorax, this case would certainly seem to support the utility of such an approach, especially in patients with a history of environmental asbestos exposure.

Learning points

- ▶ Malignant pleural mesothelioma can present in unusual ways such as spontaneous pneumothorax or hydropneumothorax.
- ▶ Consideration should be given to CT scanning in older patients presenting with spontaneous hydropneumothorax or pneumothorax to identify potential underlying malignancy, particularly when there is a history of environmental asbestos exposure.

Contributors HEF and JG had the idea of writing this case report. They were both involved in looking after the patient. HEF and VKW performed literature search and wrote the article. JG reviewed the article before submission. HEF is the guarantor for this article.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 British Thoracic Society Standards of Care Committee. BTS statement on malignant mesothelioma in the United Kingdom. *Thorax* 2007;62:ii1–19.
- 2 Saleh HZ, Fontaine E, Elsayed H. Malignant pleural mesothelioma presenting with a spontaneous hydropneumothorax: a report of 2 cases. *Rev Port Pneumol* 2012;18:93–5.
- 3 Guha K, Jones D, James HK, *et al*. Recurrent hydropneumothorax as a presenting feature of malignant mesothelioma. *Eur J Intern Med* 2008;19(Pt1):63–4.
- 4 Serap H, Hanta I, Karataş M. Spontaneous hydropneumothorax: an unusual presentation of malignant pleural mesothelioma. *Turk Resp J* 2002;3:127–9.
- 5 Wu H, Tino G, Gannon FH, *et al*. Lepidic intrapulmonary growth of malignant mesothelioma presenting as recurrent hydropneumothorax. *Hum Pathol* 1996;27:989–92

- 6 Margery J, Regnard JF, Chompret A, *et al.* Malignant mesothelioma of the pleura revealed by bilateral spontaneous pneumothorax. *Rev Pneumol Clin* 2005;61 (5 Pt 1):332–3.
- 7 Steehs N, Huizinga TW, Dik H. Bilateral hydropneumothoraces in a patient with pulmonary rheumatoid nodules during treatment with methotrexate. *Ann Rheum Dis* 2005;64:1661–2.
- 8 Daoud BT, Patel RG. Pulmonary sarcoidosis: presentation as bilateral spontaneous hydropneumothorax and pulmonary infiltrates. *Southern Med J* 2000;93/5:494–6.
- 9 Hodgson JT, McElvenny DM, Darnto AJ, *et al.* The expected burden of mesothelioma mortality in Great Britain from 2002 to 2050. *Br J Cancer* 2005;92:587–93.
- 10 MacDuff A, Arnold A, Harvey J, *et al.* Management of spontaneous pneumothorax: British Thoracic Society Pleural Disease Guideline 2010. *Thorax* 2010;65(Suppl. 2): ii18–31.
- 11 American college of Chest Physicians. Management of spontaneous pneumothorax. *Chest* 2001;119:590–602.

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow