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Unusual association of diseases/symptoms

An unusual clinical case of haemoptysis in spontaneous pneumothorax: blood clots within emphysematous bulla

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Abstract

We report an unusual case of spontaneous haemopneumothorax associated with haemoptysis due to blood clots within emphysematous bulla in a 42-year-old man. Haemoptysis disappeared after surgery.

BACKGROUND

Spontaneous pneumothorax is reported as being associated with haemoptysis in cystic fibrosis, $\underline{1}$ in congenital adenomatoid cysts of the lung, $\underline{2}$ infectious lung diseases and neoplasms. We herein report a case of haemoptysis due to the presence of blood clots in emphysematous bulla in a patient with left spontaneous haemopneumothorax.

CASE PRESENTATION

A 42-year-old man, smoker, with positive familial anamnesis of chronic obstructive pulmonary disease, was admitted urgently into our institution after 3 days of dyspnoea, acute chest pain on the left side and nightly episodes of haemoptysis.

INVESTIGATIONS

Chest *x* rays showed left hypertensive pneumothorax. The day after, *x* rays revealed a partial reexpansion of the left lower lobe and the complete collapse of the left upper lobe. Furthermore, subcutaneous emphysema had developed. Chest CT was performed and multiple emphysematous bullae, some with hyperdense area, were demonstrated in the poorly ventilated left upper lobe (fig 1). Large bullae were also seen in the contro-lateral lung (fig 1).



Figure 1

CT of the chest. Multiple bilateral emphysematous bullae, mediastinal and subcutaneous emphysema are demonstrated. Hyperdense area is showed in a bulla of the left lung.

TREATMENT

A minimally invasive left thoracic approach was performed by three ports in a triangular position, accumulated blood was removed from the chest and several bullae were seen in the upper lobe. The parietal pleura appeared thick and hyperaemic. Heavy vascularised adhesions between visceral and parietal pleura, aortic arch, succlavian artery, descending aorta and pericardium were removed. An extensive exploration allowed us to see one ruptured bulla containing blood clots (fig 2). An endoscopic linear cutter stapler was used to resect the emphysematous left upper lobe. A lung biopsy in an apparently normal lung was also carried out and localised pleurectomy was performed, from the first to the sixth intercostal space, to obtain surgical pleurodesis. Surgical sealant was applied over the stapled lines. Two chest tubes were inserted and connected to an underwater drainage system.



Video-assisted thoracic surgery. Intraoperative view. Blood clots within broken bulla.

OUTCOME AND FOLLOW-UP

The postoperative course was excellent and the drains were removed on the fourth day; the next day patient was discharged. The final histological diagnosis revealed bullous emphysema and fibrosis. The apparently normal lung showed the presence of an underlying emphysematous disease. The parietal pleura presented fibrous thickening with areas of reactive hyperplasia.

In May 2008, the patient underwent surgery to treat the emphysematous bullae on the right side and was discharged on the fourth post-operative day.

DISCUSSION

Although haemoptysis is reported as being associated with numerous lung diseases, the presence of blood clots within pulmonary emphysematous bulla causing haemoptysis in spontaneous pneumothorax is, to our knowledge, unreported in the English medical literature. The strong correlation between several episodes of haemoptysis and the presence of blood clots in the bulla is proved by the fact that these events disappeared after surgery.

The physiopathological mechanisms regarding the formation of the intrabullous blood clots can be similar to those suggested for haemothorax in spontaneous pneumothorax. Some authors, in fact, describe the presence of torn adhesions between the pleuras₃ or anomalous vascularised bullae; \pm others demonstrate congenital aberrant vessels between parietal pleura and bullae. We think that, in our patient, the vascularised adhesions between the bulla and the parietal pleura were torn and started to bleed inside and outside the bulla.

A CT of the chest is in our institution always carried out when, after inserting a chest drain, lung reexpansion is delayed or air leakage is persistent. As regards to our patient, the presence of a hyperdense area within the bulla suggested the presence of blood clots, which have been confirmed intraoperatively.

Haemoptysis in patients with spontaneous pneumothorax and bullous disease indicates the presence of blood clots within the bullae and can require an immediate surgical intervention to avoid serious complication such as empyema or lung abscess.

LEARNING POINTS

- Haemoptysis during spontaneous pneumothorax may indicate presence of haemothorax and airways flooding via the bronchopleural laceration that caused the pneumothorax.
- Immediate surgery is indicated to avoid serious complications such as empyema or lung abscess.

Footnotes

Competing interests: none. Patient consent: Patient/guardian consent was obtained for publication.

REFERENCES

1. Flume PA, Strange C, YE X, et al. Pneumothorax in cystic fibrosis. Chest 2005; 128: 720–8.[PubMed]

2. Lee SC, Cheng YL, YU CP. Haemopneumothorax from congenital cystic adenomatoid malformation in a cryptorchidism patient. Europ Resp J 2000; 15: 430–2.

3. Tatebe S, Kanazawa H, Yamazaki Y. Spntaneous hemopneumothorax. Ann Thorac Surg 1996; 62: 1011–15. [PubMed]

4. Rowell NR. Spontaneous haemopneumothorax. Br J Tuberc 1956; 50: 214-20.

5. Muragushi T, Tsukioka K, Hirata S, et al. Spontaneous hemopneumothorax with aberrant vessels found to be the source of bleeding: report of two cases. Surg Today 1993; 23: 1119–23. [PubMed]