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Case Study

An Uncommon Vascular Cause of Spontaneous Hydropneumothorax

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ABSTRACT

This paper highlights a potentially life threatening and unsuspected case of multiple small pseudoaneurysms from the thyrocervical and costocervical branch of the left subclavian artery that was spontaneously ruptured. The cause was not suspected prior to thoracic CT angiography (CTA) because the initial pre-emptive diagnosis was only trivial spontaneous hydropneumothorax which became serious after a chest tube was inserted.

Keywords: Hydropneumothorax, vascular

INTRODUCTION

Conventional CT thorax is a known method to diagnose non-vascular lung pathology, while CT angiography (CTA) is a special technique in image acquisition used mainly to visualize the vascular structure. In this study CTA was used to rule out any iatrogenic injury to the vessel post chest tube insertion. This method enabled us to promptly diagnose pseudoaneurysm as the cause of spontaneous hydropneumothorax (SHP). The Conventional CT thorax would probably miss the cause of SHP and further delay diagnosis and treatment.

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SHP can be due to thoracic malignancies, vascular malformations, infections, coagulation disorders or cavitating pulmonary infarction (Suppiah & Abdullah, 2015). SHP has high mortality due to rapid blood loss and aerated lung volume loss. The incidence of SHP is up to 7% of patients with spontaneous pneumothorax (Ali, Lippmann, Mundathaje & Khaleeq G, 2008, Hsu, Shih, Hsu, & Chen, 2005). SHP usually occurs with the presence of illness or injury. Anomalous vascular cause for SHP is extremely rare. The aetiology can be varied but includes lung neoplasms, blood dyscrasia, arteriovenous malformation (Osler-Weber Rendu Disease), pregnancy, aortic dissection, pulmonary emboli, connective tissue disorders such as Ehlers-Danlos syndrome (EDS) type IV, endometriosis, adhesions with pneumothorax and infection (Hsu et al, 2005). SPH has a predilection among males with 100% male dominance. Smokers are thought to have a higher incidence of SHP accounting for 76.5% of patients (Kim, Kang, Pyo, Jeon, & Lee, 2008).

RESULT

A 21-year-old male, social smoker with no underlying medical illness was presented to the Emergency Department (ED) with a sudden onset of dyspnoea associated with epigastric pain radiating to the back. Upon examination, he had severe, left-side chest pain. No history of trauma or heavy coughing was identified. A Chest X-ray (CXR) (Figure 1A) confirmed a left hydropneumothorax with tracheal deviation to the right. An intercostal chest drain was inserted and approximately 1.7 litres of haemoserous fluid was drained over three hours.

The patient became progressively hypotensive with a haemoglobin level of 6.8 g/dL and Haematocrit of 19%. An urgent non-cardiac gated CT-angiography with 128-DECT (dual energy) CT scanner (Somatom Sensation Flash, Siemens Medical Solutions) was immediately performed to exclude any iatrogenic chest drain-related lung injury. The CTA demonstrated a left hydropneumothorax with an active contrast extravasation into the left pleural space that was suggestive of active bleeding (Figure 1B). The actual origin of the haemorrhage was not well delineated on the CTA. However, a well-defined lobulated arterial density structure was seen in the upper lobe suggesting pseudoaneurysm (Figure 2 A, 2B). No active contrast extravasation was noted around the chest drain site. The patient was transferred to the Angiography suite from red zone ED. The interventional radiology team swiftly proceeded with an angiography using the right femoral puncture approach. The left subclavian artery (SCA) digital subtraction images revealed a leaking pseudoaneurysm supplied from the thyrocervical branch of the left SCA (Figure 3A, B and C). The feeder vessel of the thyrocervical pseudoaneurysm branch was embolized using Polyvinyl Alcohol (PVA) particles, 250-355 μm size and a single 2.0 mm x 20.0 mm coil. The subsequent post-embolization run of the left SCA demonstrated another, smaller leaking pseudoaneurysm from the costocervical branch of left subclavian artery (Figure 3D), which was embolized with PVA particles 250-355 μm.

The post-embolisation subsequent angiogram showed complete resolution of contrast extravasation. The patient's condition steadily improved upon receiving a total of 4 pints of pack red blood cells (RBC) for resuscitation. Serial CXR's showed improvement of the left hemopneumothorax allowing the patient to be discharged a week later.

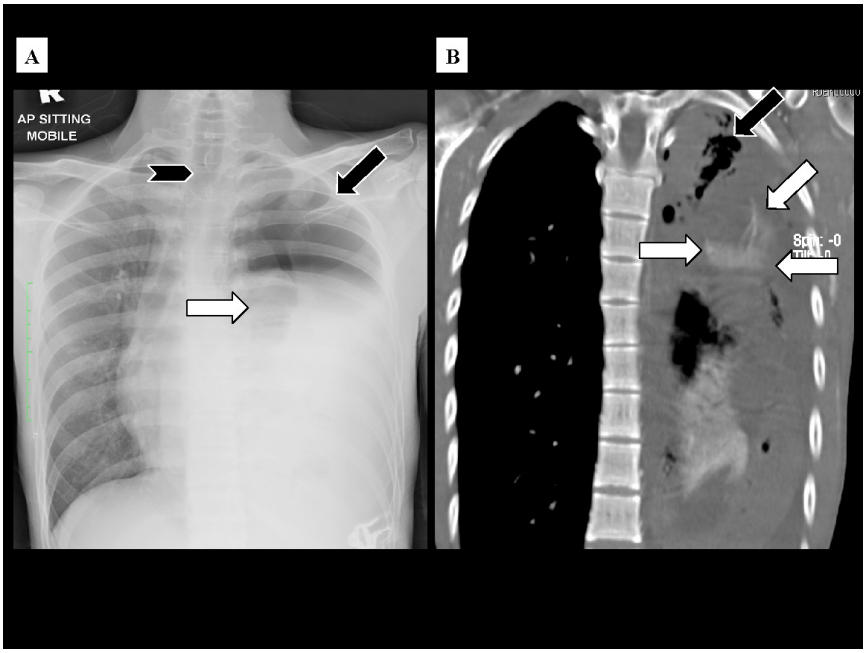

Figure 1. CXR in casualty showed left hydropneumothorax; pneumothorax (black arrow) with fluid level within (white arrow) and tracheal deviation to the right (notched arrow) (A). Coronal CT Thorax image in delayed images showing pooling of contrast in the left upper hemithorax (white arrow) and air pockets within the effusion confirming the pneumothorax component (black arrow) (B)

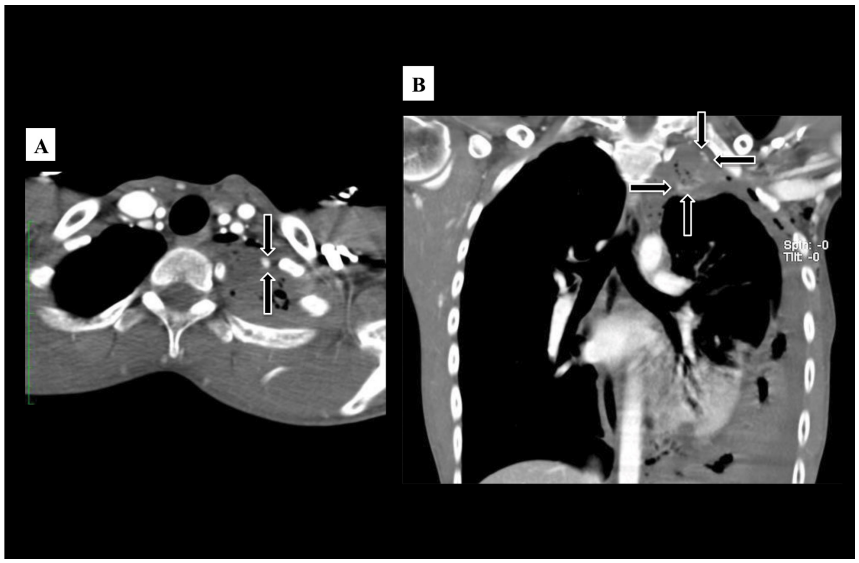

Figure 2. Axial CTA Thorax image in arterial phase showing the lobulated enhancing structure in apical region (black arrow) suggestive of a pseudoaneurysm (A). Coronal CT Thorax image in arterial phase showing active contrast extravasation in the left apical region (black arrow) (B)

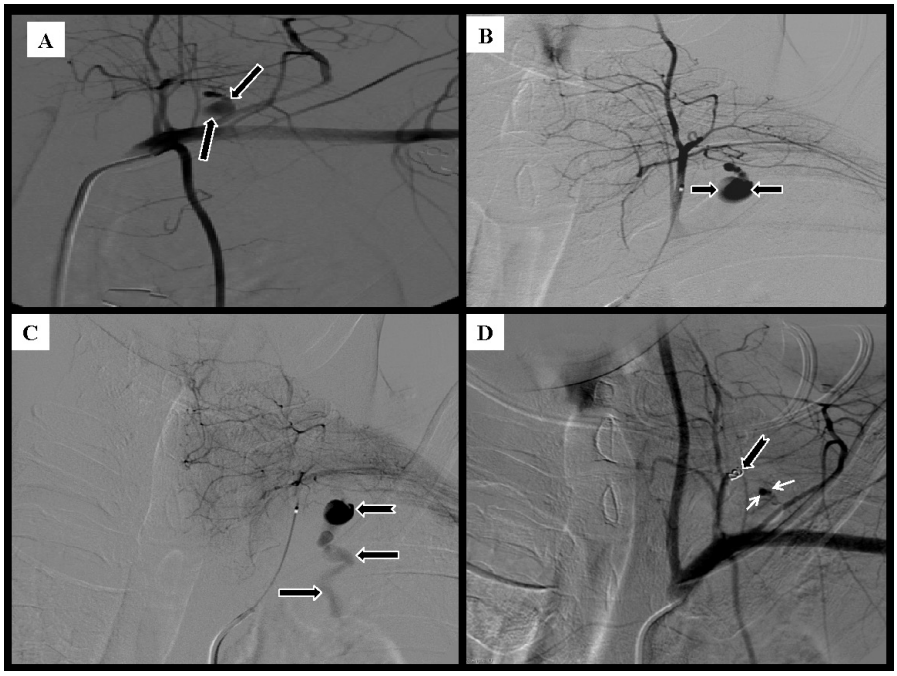

Figure 3. Left SCA angiogram showing pseudoaneurysm (black arrow) (A). Selective angiogram in left thyrocervical trunk confirms pseudoaneurysm (black arrow) (B). Pseudoaneurysm (notch arrow) with active extravasation of contrast into left hemithorax (black arrow) (C). Post-coiling run showing coil in situ (notch arrow) and left costocervical trunk pseudoaneurysm (thin white arrow) (D)

DISCUSSION

The cause of haemothorax is hypothesized due to a normal vessel having undergone chronic stress, due to abnormal vascular morphology or due to torn adhesion of the lung pleura or even rupturing of the luxury perfused bullae (Ali et al., 2008; Kanazawa, Yamazaki, Aoki, & Sakurai, 1996). Conditions such as coagulopathy, neoplasia, and endometriosis are also a few of the causes (Patrini et al., 2015). Mucoid degeneration within the aberrant vessel wall was also reported to be one of the causes of SHP (Tatebe et al., 1996). In our patient, there was no evidence the above-mentioned conditions, coagulopathy or trauma. In such circumstances, spontaneous rupture of intrathoracic blood vessels needs to be considered.

Despite the initial CXR, which demonstrated a hemopneumothorax, the bleeding source in our patient was identified during angiography arising from a pseudoaneurysm supplied by the thyrocervical and costocervical branches of the left Subclavian artery (SCA). The thyrocervical arterial branch that originates from SCA is well protected from blunt trauma due to its deep bodily location. It is most vulnerable during central venous catheterization. Our patient had no history of any previous attempts at central venous access.

There is a long list for the causes of pseudoaneurysm, such as direct trauma or indirect trauma to the vessel wall, disease vessel wall, vasculitides, dissections and even inflammation. Pseudoaneurysm happens when there is a defect in the vessel wall with an intact adventitia,

causing blood to leak through the defect, forming a sac that is only lined by the tunica adventitia. Due to its poor wall strength, the risk of rupture is significantly greater (Marx, Gardiner, & Miller, 1985).

An uncommon correlation between neurofibromatosis (NF) and arterial aneurysm due to wall fragility was also reported in patients with SHP and the incidence is approximately 3.6% (Salyer & Salyer, 1974). In regards of NF, the bleeding is probably either due to rupture of the abnormal vessels or bleeding schwannoma. Bleeding from the thyrocervical trunk in NF type 1 has been reported, although it is not common compared with bleeding from SCA and intercostal arteries which is more common in NF type 1 (Salyer & Salyer, 1974; Miura et al., 1997). Our patient had no features suggestive of NF.

Historically, surgery has been the main stay of treatment for SHP; however, the trend for less invasive methods such as the video-assisted thoracoscopic (VATS) approach has become popular. Endovascular treatment is also effective and its increasing availability in tertiary care centres all over the world has made it a good alternative. Coils embolization is the treatment of choice in the end vessels whereby the small artery feeding the pseudoaneurysm can be sacrificed. A covered stent is the best choice to control arterial bleeding in pseudoaneurysm that arises from the main parent artery. In our patient, the bleeding site was a ruptured pseudoaneurysm from the small thyrocervical branch and costocervical arteries, hence coil embolization was utilized.

CONCLUSION

SHP is potentially life threatening. It can result in massive blood loss, severe respiratory compromise and mortality if not promptly treated. A thoracotomy however is not always indicated, as has been the case in the past. A contemporary endovascular approach with coils embolization may be technically feasible being less invasive and safer than open surgery. However, had this patient continued to bleed post-embolization or if the latter failed, emergency surgical exploration would have been necessary. Similarly, surgery is more expedient for a hemodynamically unstable patient. Hence close collaboration between the interventional radiologist and cardiothoracic surgeon is vital to ensure optimal care.

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REFERENCES

Ali, H. A., Lippmann, M., Mundathaje, U., & Khaleeq, G. (2008). Spontaneous hemothorax: a comprehensive review. *Chest Journal*, *134*(5), 1056-65.

Hsu, N. Y., Shih, C. S., Hsu, C. P., & Chen, P. R. (2005). Spontaneous hemopneumothorax revisited: clinical approach and systemic review of the literature. *The Annals of Thoracic Surgery*, *80*(5), 1859-63.

Kim, E. S., Kang, J. Y., Pyo, C. H., Jeon, E. Y., & Lee, W. B. (2008). 12-year experience of spontaneous hemopneumothorax. *Annals of Thoracic and Cardiovascular Surgery*, *14*(3), 149-153.

Marx, M., Gardiner, G. A. Jr., & Miller, R. H. (1985). The truth about false aneurysms. *American Journal of Roentgenology*, *145*(1), 193-194.

Miura, H., Taira, O., Uchida, O., Usuda, J., Hirai, S., & Kato, H. (1997). Spontaneous haemothorax associated with von Recklinghausen's disease: review of occurrence in Japan. *Thorax*, *52*(6), 577-578.

Patrini, D., Panagiotopoulos, N., Pararajasingham, J., Gvinianidze, L., Iqbal, Y., & Lawrence D. R. (2015). Etiology and management of spontaneous haemothorax. *Journal of Thoracic Disease*, *7*(3), 520–526.

Salyer, W. R., & Salyer, D. C., (1974). The vascular lesions of neurofibromatosis. *Angiology*, *25*(8), 510-519.

Suppiah, S., & Abdullah, B. J. J. (2015). Spontaneous haemopneumothorax due to cavitating pulmonary infarction - a rare condition revisited. *International Journal of Public Health and Clinical Sciences*, *2*(2), 45 -54.

Tatebe, S., Kanazawa, H., Yamazaki, Y., Aoki, E., & Sakurai, Y. (1996). Spontaneous hemopneumothorax. *The Annals of thoracic surgery*, *62*(4), 1011-1015.