

Incidental diagnosis and therapeutic approach of an iatrogenic intra-parenchymal pulmonary intercostal artery pseudoaneurysm: a case report

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Abstract

Intercostal artery pseudoaneurysms (IAP) are extremely rare but its sudden rupture represents a life-threatening complication. We report an unusual case of a late intercostal artery pseudoaneurysm, after a video-assisted thoracoscopic surgery, presenting as a large intra-parenchymal lung mass. Diagnosis was made by biphasic computed tomography angiography and contrast-enhanced pulsed-wave doppler ultrasound. To minimize the risk of aneurysm bleeding immediate angiographic embolization was planned and successfully performed. IAP should be considered in presence of lung mass in patients who have previously undergone an intervention with intercostal approach to prevent to prevent life-threatening complications.

Introduction

Intercostal artery pseudoaneurysm (IAP) is a rare complication of surgical procedures involving intercostal space. It is generally

asymptomatic and difficult to diagnose until its rupture. Sudden bleeding may occur in IAP, generally into pleural space or retroperitoneum, with a rapid decline in cardiopulmonary status, with high risk of mortality in absence of treatment [1,2].

In presence of comorbidity, symptoms may be aspecific reflecting different phenotypes of underlying diseases [3-7]. We present the first report in literature of a late onset intercostal artery pseudo aneurysm following video-assisted thoracoscopic surgery (VATS), presenting as large intra-parenchymal mass. Among diagnostic procedures imaging is relevant for differential diagnosis of lung intra-parenchyma mass and CT angiography is crucial to characterize vascular involvement [8-12]. Following diagnosis by biphasic computed tomography angiography and contrast-enhanced pulsed-wave doppler ultrasound study, an immediate angiographic embolization was planned and successfully performed to minimize the risk of aneurysm bleeding.

Case Report

An 82-year-old male, ex-smoker with asbestos exposure, with clinical history of pacemaker implantation for trifascicular heart block in 2016 and COPD presented with one month of exertional dyspnoea unresponsive to pharmacological treatments. He had clinical history of pleuritis in fibrotic evolution, diagnosed by VATS biopsy performed during diagnostic procedure for parenchymal consolidation associated with pleural effusion in 2014.

The Chest X-Ray (CXR) revealed a large opacity in the right lower lobe. A Chest Computed Tomography (CT) was performed, showing presence of a non-homogeneous well capsulated pulmonary mass (6x5.5 cm) (Figure 1a). No alteration in blood chemistry and a mild reduction of alveolar capillary diffusion capacity was observed. A percutaneous CT-guided fine-needle aspiration biopsy (FNAB) detected the presence of haematic material. Following radiologic case review, a biphasic CT angiography was performed; the pre-surgical chest CT was analysed and a tiny colligation nuclei in the fibrotic collapsed parenchyma adjacent to the pleural chronic thickening was identified (Figure 1b). A biphasic CT angiography was then performed on pulmonary and systemic vessel which showed the vascular nature of the mass: a network of tubular vessels, surrounded by extensive clot and hyperdense walls. The diagnosis of a chronic pseudo-aneurysm was finally made (Figure 1 c,d). To determine the systemic or pulmonary feeding of the mass, a

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contrast-enhanced pulsed-wave doppler ultrasound study (PWDUS) was performed, which confirmed the prevalence of systemic origin of the vessels (Figure 2 a,b). Patient underwent digital subtraction angiography (DSA) that detailed the presence of a pseudoaneurysm arising from XII right intercostal artery and draining into the inferior lobar vein (Figure 2c). The embolization of the XII right intercostal artery was performed, across femoral access, using two coils of Microplex10 5/22 mm and 5/6 mm, respectively. There were no complications in the execution of the embolization and in post-operative period. Post embolization angiography showed successful occlusion of the pseudoaneurysm (Figure 2d).

After one month, the patient reported an improvement in respiratory symptoms and CT angiography examination showed a marked reduction of intralesional vessels. Follow-up visits continued with functional examinations and Tc-scan that showed stability in both the respiratory symptoms and radiological scan, up to the last 2-year check-up from the interventional procedure.

Discussion

A growing body of researches reported that VATS and minimally invasive surgical techniques have a worthy safety profile in both benign and malignant thoracic diseases [13-16]. IAP is extremely rare with only few cases reported in the literature to our knowledge. The aetiology has been reported to be traumatic in two [17,18] and iatrogenic in eleven cases due to procedures involving an intercostal approach [1,2,19-21]. VATS procedure was identified cause of IAP only in one case in literature [19]. In our case the lesion vessel appeared to emerge very close to pleural biopsy site. We can hypothesize that the injury was made by contact of the VATS instruments or drainage tubes with visceral pleural vessels. The late onset of the pseudoaneurysm formation is confirmed by a negative post-operative CT scan performed nine months after VATS. In most of the cases IAP presented within one month of the procedure. We incidentally diagnosed IAP two years

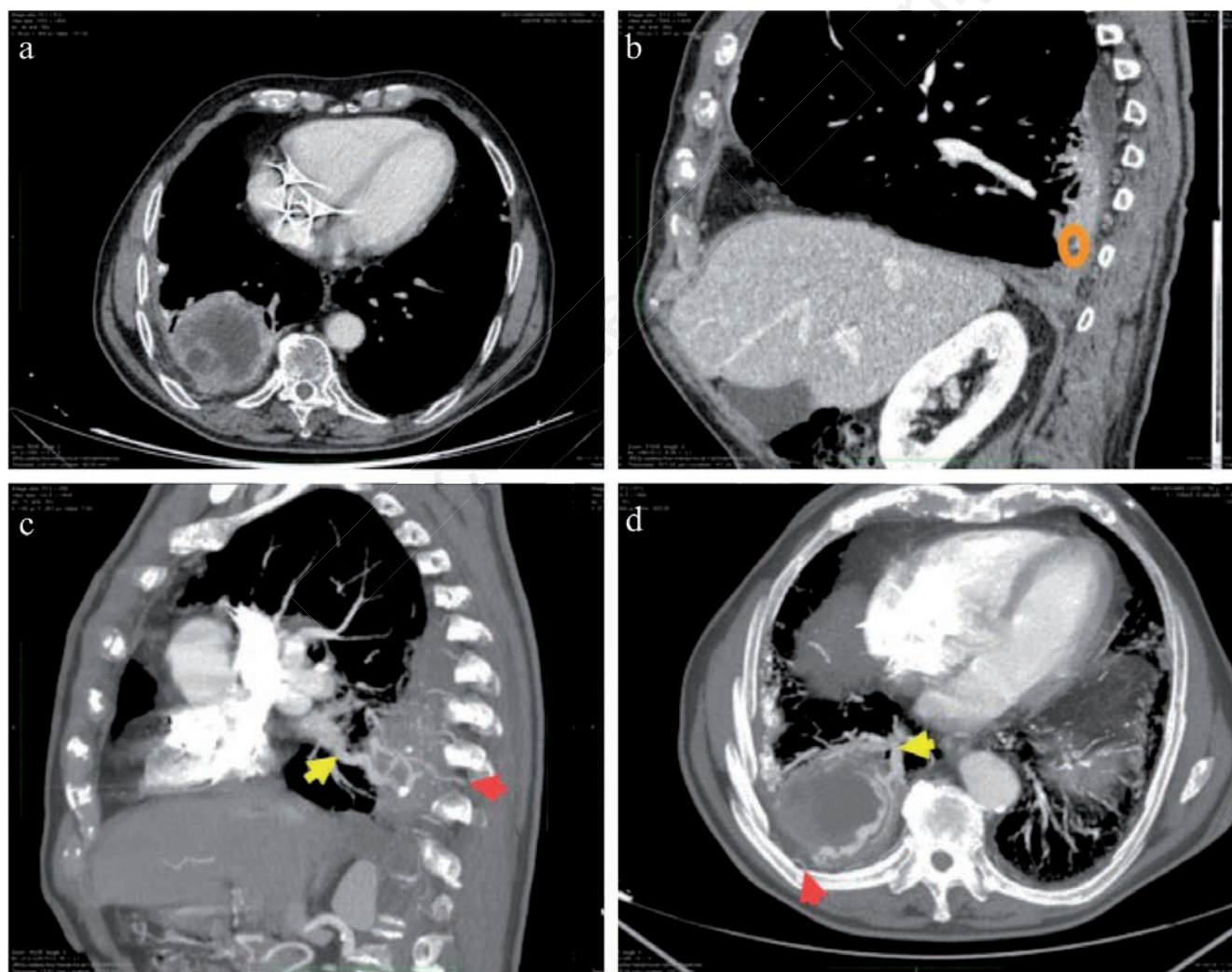


Figure 1. a) Axial contrast-enhanced CT scan. Heterogeneous well capsulated pulmonary mass is seen in inferior right lobe (6x5.5 cm). b) Sagittal chest CT shows parenchyma consolidation with proximal air bronchogram, tiny calcified parenchyma and pleural nodule and a colliquative intra-parenchyma fluid loculation (orange circle) and organized pleural effusion is also evident. c,d) Sagittal and Axial Maximum Intensity Projection (MIP) of the CT Angiography shows a network of tubular vessels, rounded by extensive cloth of and hyperdense wall: XII intercostal artery (red arrow) and pulmonary vein (yellow arrow) are evident.

after the intervention of VATS, during investigations for exertional dyspnoea.

Haemothorax, due to the bleeding of pseudoaneurysm into pleural space, is the most common clinical presentation; atypical onset with a pulsatile mass [19,22], an extrapleural hematoma [23] and a retroperitoneal haematoma [24] have also been reported in literature.

To our knowledge this is the first case of large tumour-like intraparenchymal mass presentation of the pseudoaneurysm. Our patient presented with exertional dyspnoea in part due to underlying COPD without haemoptysis nor fatigue.

Due to the risk of early bleeding after its formation, diagnosis and treatment before rupture is critical. Definitive diagnosis of IAP is usually made by DSA [20-22], but CT and US can be very useful as primary diagnostic modality [21,22,25]. In our case, a biphasic CT angiography on pulmonary and systemic vessel was fundamental to recognize the vascular nature of the mass and make the diagnosis. The imaging showed the typical radiological findings of intraparenchymal IAP: a network of tubular vessels surrounded by

extensive clot and hyperdense walls of collapsed lung. Furthermore, we used contrast-enhanced PWDUS for the first time on IAP to confirm the systemic origin of feeding vessels and guide the therapy.

Embolization with metallic or microcoils is considered the first line treatment [26]. There are few reports of treatment with covered stents [27], percutaneous thrombin injections [28], post embolization [21] or aneurismectomy [27] as first line of treatment and two cases of conservative management [25,28]. We successfully treated our patient with embolization of the XII right intercostal artery using two coils of Microplex100 5/22 mm and 5/6 mm, respectively.

Conclusions

Intercostal artery pseudoaneurysm should be considered in presence of lung mass in patients who have previously undergone

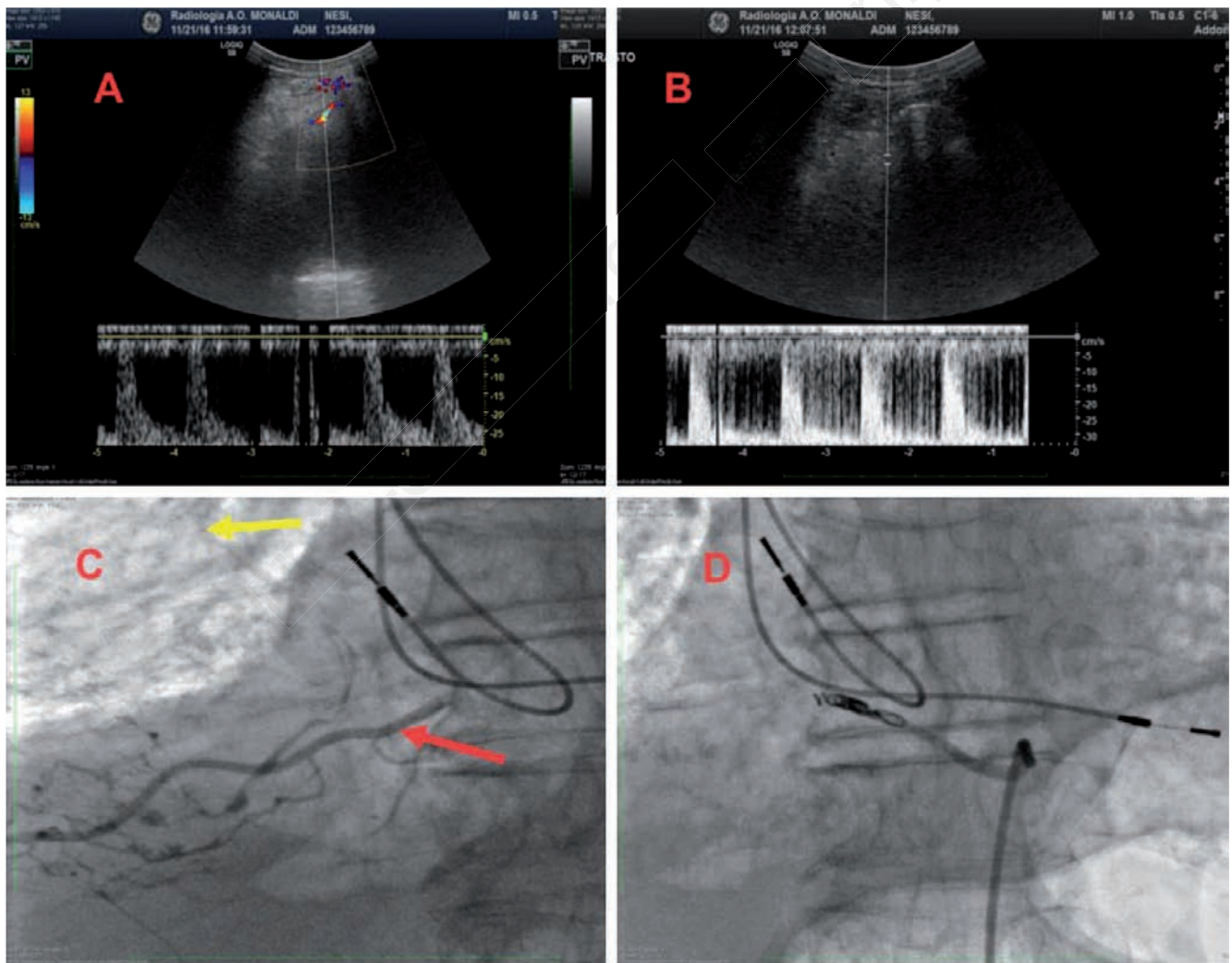


Figure 2. a) Ultrasound study of the pseudo-aneurysm feeding vessel; the color Doppler ultrasound hardly shows the signal of the feeding vessel. b) The contrast-enhanced power wave Doppler ultrasound clearly shows the typical systemic spectral feature of vessel; c) Digital subtraction angiography (DSA); the DSA, due to its better spatial resolution, clearly evidences the thin network of intraparenchyma vessels (red arrow); in the static imagine it is hardly evident the draining pulmonary vein (yellow arrow). d) Post-embolization DSA evidences absence of flow of the intercostal feeding artery distal to micro-coils.

an intervention with intercostal approach. CT angiography is essential to the diagnosis in these patients and contrast-enhanced PWDUS can guide the subsequent minimally invasive treatment. Early angiographic treatment should be carried out as emergency procedure after diagnosis of IAP to prevent life-threatening complications.

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