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Case report

Trancatheter embolization of pulmonary artery pseudoaneurysm with detachable coils in association with non-adhesive liquid embolizing agent (Squid)

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Abstract. A nodular lesion at the lower left pulmonary lobe was detected in a 46 years old male during a preoperative chest X-Ray for appendicitis. To further characterise the nodule, a contrast enhanced computed tomography (CE-CT) was performed showing a 20 mm vascular lesion, which was suspected to be a pseudoaneurysm. The diagnostic angiography detected a flattening of the vascular wall with a voluminous pseudoaneurysm (PSA) in the distal portion of the tributary branch of the inferior left lobe. To treat the lesion, a 2.9F microcatheter was advanced into the sac and 4 detachable coils were placed (16-18mm Penumbra Inc) to pack the PSA. To block vascular supply to the lesion, the feeding artery was embolized with an ethylene vinyl alcohol copolymer agent (Squid Peri 18, Emboflu). The final angiographic control showed the exclusion of the pseudoaneurysmal sac which was confirmed by SEMAR™ reconstructed CE-CT scan after 40 days. Furthermore, no signs of pulmonary infarction were reported. (www.actabiomedica.it)

Key words: aneurysm, angiography, CT angiography, embolization, interventional

Introduction

Pulmonary artery pseudoaneurysms (PAPs) are rare but often lethal, this vascular pathology could be found incidentally on radiological examination or could begin with massive hemoptysis.

Among the causes of PAPs such as infections, iatrogenic traumas and malignancies: PAPs are commonly associated with Swan-Ganz catheter placement. In the literature, approximately 97% of iatrogenic pulmonary pseudoaneurysms are described in the right pulmonary circulation.

Here we described the diagnosis and treatment of a PAPs in the left lower lung lobe, incidentally found during a routine investigation.

Case Report

A 46 years old moroccan male, at the preoperative chest X-ray for an acute phlegmonous appendicitis was found to have a nodular hyperdiafanic lesion at the lower left pulmonary lobe (Figure. 1 a). To obtain a more complete characterization of the lesion, a contrast enhanced computed tomography (CE-CT) (Aquilion Prime SP, Canon Medical, The Netherlands) was performed (Figure. 1a, 1b) showing a lesion of 20 mm (Figure. 1 b).

This formation had a contrast enhancement equal to that of the pulmonary arteries. Moreover, by performing targeted reconstruction (maximum intensity projection (MIP) on the paracoronal plane) (Figure. 2 a) a communication tract between the lesion and a tributary

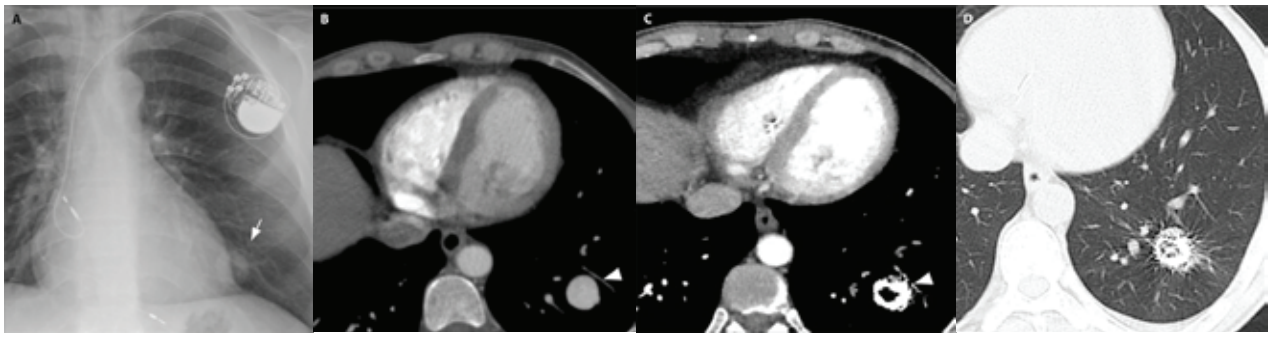

Figure 1. a) Preoperative Chest X ray shows a nodular radiopaque lesion at the left lower lobe (white arrow). Associated with this finding no pulmonary consolidation of a phlogistic or dysventilatory appearance were recognised. b) CE-CT on the axial plane demonstrates a saccular dilation of the pulmonary artery tributary of the posterior segment of the inferior left lobe (white arrowhead). The density measured in Hounsfield unit within the lesion was equal to that measured within the pulmonary arterial vessels, allowing us to hypothesise that there was communication between the lesion and the pulmonary circulation. The absence of pathological findings in the adjacent lung parenchyma was also confirmed. c) CE-CT on the axial plane, using SEMAR™ software at 1 month after the procedure confirmed the complete embolization of the vascular lesion: coils are leaning on the wall of the lesion without iodized contrast medium in the core (arrowhead). SEMAR™ significantly reduced beam hardening artefacts so that revascularization of the lesion could be excluded with certainty. d) CE- CT on the axial plane, using SEMAR™ software at 1 month after the procedure, (lung window), neither demonstrates no signs of pulmonary infarction in the lung parenchyma surrounding the coils or in the more peripheral subpleural areas.

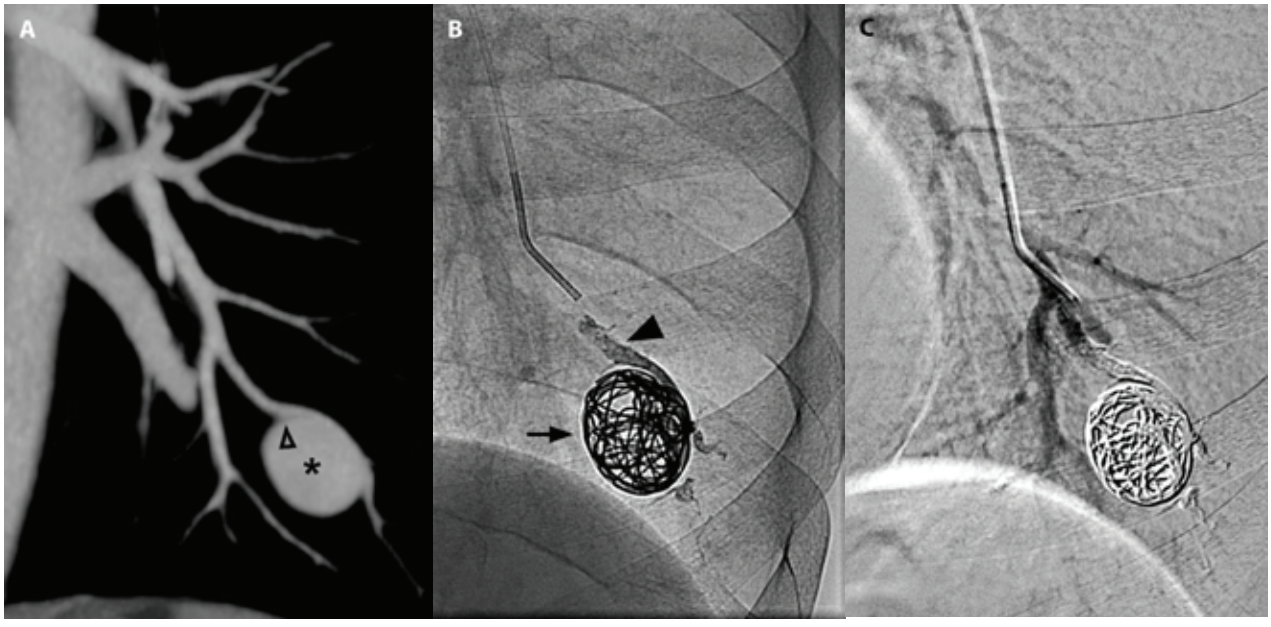

Figure 2. a) CE-CT maximum intensity projection (MIP) on the paracoronal plane demonstrates the saccular dilation of the pulmonary artery tributary of the posterior segment of the left pulmonary lobe (asterisk). A septum of the vascular wall can be observed, which divides the lumen of the afferent vessel from the lesion sac (empty arrowhead). b) Post procedural fluoroscopy control demonstrates distribution of the radiopaque embolizing liquid inside the tributary vessel (black arrowhead) and the complete occupation of the vascular pseudoaneurysmatic sac by detachable coils (arrow). c) Post procedural digital subtraction angiography (DSA) shows the embolization of the pulmonary artery tributary of the posterior segment of the left pulmonary lobe with no contrast medium inside the PSA.

arterial branch to the left lower pulmonary lobe was identifiable. In addition, in the cranial part of the lesion, a septum of the vascular wall can be observed, which divides the lumen of the afferent vessel from the lesion sac (Figure. 2 a). This detail was crucial in distinguishing the lesion described here from a "true" aneurysm.

Aneurysms, by definition, are formed by the entire wall of the arterial vessel, whereas in this case only the tunica media and adventitia constitute the vascular lesion. Furthermore, in the Literature, there are no cases describing "true" aneurysms of the distal pulmonary arteries. No signs of rupture of the vascular lesion were reported, such as the presence of extravasation of contrast medium in the lung parenchyma. The patient, 10 years before, suffered from a complete atrioventricular block that needed a temporary pacemaker, positioned through a femoral access; subsequently a definitive one was placed. No complications were reported during the procedure and the patient didn't suffer from pulmonary symptoms over the annual cardiological follow up. The above mentioned findings, in association with the history of a previous venous catheterisation, led to a diagnosis of pulmonary pseudoaneurysm. According to the preoperative findings, after a multidisciplinary consultation, it was decided to candidate the patient for a percutaneous embolization procedure.

The treatment was performed in the angiographic-suite with monitoring of vital parameters without any type of sedation, only local anesthesia. Through the right femoral vein the left pulmonary artery was reached with a 5-French vertebral catheter (Cordis,Miami, FL, USA). Subsequently a diagnostic angiography showed a flattening of the vascular wall conditioning a voluminous pseudoaneurysm in the distal portion of the tributary branch of the left lower lobe.

To perform the selective embolization, a 2.9F microcatheter (Terumo, Progreat, Tokyo, Japan) was advanced in the tributary vessel and through it, 4 detachable coils were landed inside the sac (16-18mm Penumbra Inc; Alameda, USA). To better exclude the lesion from its vascular supply, the feeding artery was embolized with one vial of an ethylene vinyl alcohol copolymer (EVOH) agent (Squid Peri 18, Emboflu, Gland, Switzerland). Before injecting the embolic fluid, the dead space of the microcatheter was filled with 0.4 mL of dimethyl sulfoxide (DMSO) solvent then Squid was slowly injected in about 60 seconds; when the Squid started flowing back to the proximal part of the feeding vessel, the microcatheter was pulled out (Figure. 2 b).

The final angiographic control showed the complete exclusion of the pseudoaneurysmal sac; this finding was further confirmed by the control CE-CT after 40 days (Figure. 1 c - Figure. 2 c). No signs of pulmonary infarction were reported at the control CE-CT (Figure. 1 d)

Discussion

Pulmonary artery pseudoaneurysms (PAPs) are rare but can be fatal; this vascular pathology might be found incidentally on radiological examination or might start with massive haemoptysis. Furthermore, as stated by Kalra-Lall, PAPs are so rare that they are often not included in differential diagnoses and are often missed at initial imaging workup[1]. The main causes of PAPs are infection, iatrogenic trauma and malignancy. PAPs are commonly associated with Swan-Ganz catheter placement with a complication rate of 0.2% for pulmonary artery rupture[1]. The management of PAPs has changed in recent years from a wait-and-see approach to an interventional one. Currently, the endovascular approach is preferred over surgery, which used to be the first choice; now, it is only used in cases of large vascular lesions or at high risk of bleeding from an acute rupture[1]. The most commonly used embolic agent in PAPs embolisation are coils; other options include plugs, stents, particles, thrombin and liquid adhesive embolic agent (such as glue). Coils are associated with an intraoperative technical success rate of 89%[2]; these post-procedural angiographic findings are sometimes associated with revascularization of the sac, which is seen at follow-up CE-CT. Moreover, the re-intervention rate in these patients is about 50%[2]. Covered stent could have been a viable alternative in this case, as it would have preserved distal blood flow while excluding vascular lesion. However, covered stents are associated with the risk of occlusion and migration. Because of the peripherality of the lesion, its embolisation would not have caused massive pulmonary infarction. For these reasons, in this case, we chose to use a non-adhesive embolic agent (Squid) combined with the coils to maximise the embolizing power and reduce the risk of revascularization; this combination creates a more stable plug than that provided by the coils alone, as also recently reported in splenic artery aneurysms[3]. In addition, no pulmonary

infarction was found at post-procedural CE-CT. Non-adhesive embolising fluids (Squid, Onyx) compared to adhesive ones have multiple advantages: excellent embolising capacity, could reach vessels with a diameter of 80 microns with a low tendency to induce off-target embolisation and also the risk of microcatheter entrapment is lower[4]. On the other hand, the main disadvantages are the need for compatible microcatheters, their higher cost and also the mandatory use of DMSO. DMSO is mandatory to keep the embolic fluid dissolved before injection nevertheless it can cause vasospasm, endothelial wall damage and pain. Squid and Onyx as embolic agents are widely used in the interventional brain field, however recently several articles regarding their application in the extracranial district have been reported[4].

Comparing the two non-adhesive embolic fluids, Squid has 30% less tantalum and those particles are smaller in size compared to Onyx, these characteristics allow a wider distribution and decrease beam hardening artifacts. Those CT artifacts could cause severe limitations in discriminating the efficacy of the embolization procedure. In our case, the use of Squid in association with a CE-CT reconstructed by SEMAR software (single-energy iterative retrospective metal artifact reduction software that reduced near-field artifacts generated by both high- and low-density implants) has allowed us to minimize beam hardening artifacts[5] and give us the certainty to exclude a post-operative revascularization in the core of the lesion (Figure. 1c). Moreover, no signs of pulmonary infarction were found on postoperative CT (Figure. 1 d).

Analysing retrospectively the clinical history of the patient, we suspect that this vascular lesion was caused during the previous cardiac catheterisation, ten years before, even if it was asymptomatic over those years.

The patient is currently undergoing close cardiological and pulmonary clinical follow-up. In addition, after a multidisciplinary discussion with the thoracic surgeons, it was decided, in order to reduce the exposure to radiation and the administration of iodinated contrast medium, to schedule a CTA check-up approximately 12 months after surgery, in case of no preterm complications.

To our knowledge, no cases of PAPs embolization using a recent ethylene vinyl alcohol (EVOH) copolymer agent (Squid) in association with detachable coils have been previously mentioned in literature. In our preliminary experience, association of coils and Squid in this unusual vascular district was efficient in PAP embolization without complications. However further studies with a larger cohort of patients and with a longer follow-up will be necessary to support our preliminary conclusions regarding the association of Squid and coils in PAPs embolization.

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