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Case Report

Transcatheter coil embolization of a complex pulmonary artery pseudoaneurysm with thyrocervical trunk-pulmonary arterial fistulization in a patient with cystic fibrosis and massive hemoptysis ☆,☆☆

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ABSTRACT

Pulmonary artery pseudoaneurysm (PAP) is a rare cause of life-threatening hemoptysis and tends to develop in the setting of infection, neoplasm, or trauma. Successful endovascular coil embolization has demonstrated effectiveness in treating PAPs and is now the treatment of choice for these patients. Vascular supply to PAPs is highly variable and often requires embolization of both the systemic and pulmonary feeding vessels. This is a case report of a successful transcatheter coil embolization of a complex PAP with a thyrocervical trunk-pulmonary arterial fistula in a patient with massive hemoptysis in the setting of advanced cystic fibrosis.

Introduction

Pulmonary artery pseudoaneurysms (PAP), also known as Rasmussen aneurysms, tend to develop in the setting of infection, neoplasm, or trauma, with most having a propensity to form in the segmental or subsegmental pulmonary arteries [1]. Studies have demonstrated the prevalence of PAP in patients with hemoptysis who underwent angiographic studies to be between 5%-11% [1–3]. Successful endovascular transcatheter coil embolization has demonstrated effectiveness in treating PAPs and is now commonly utilized as the initial treatment for these patients [1,4-7]. Vascular supply to PAPs is often highly variable and frequently require embolization of both the systemic and pulmonary arteries supplying the pseudoaneurysm [6,8]. In this report, we present a case of success-

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ful embolization of a complex Type C PAP with a thyrocervical trunk-pulmonary artery fistula in a patient with massive hemoptysis and cystic fibrosis.

Case summary

A 32-year old female with a past medical history of cystic fibrosis (CF) complicated by bronchiectasis, recurrent hemoptysis, pancreatic insufficiency, and malnutrition was admitted for massive hemoptysis. She was initially treated with broad-spectrum antibiotics for presumed CF exacerbation. Computed tomography angiogram (CTA) of the chest revealed centrilobular ground-glass opacities in the left lower lobe, which were thought to reflect pulmonary hemorrhage. The patient was then referred to interventional radiology (IR) for bronchial artery embolization. Right common femoral artery access was obtained, and multiple diagnostic catheter combinations were used to select the left bronchial artery. Subsequent left bronchial arteriogram was performed and a microcatheter was then used to select the left bronchial artery more distally beyond the origin of a downward directed superior phrenic artery branch. The left bronchial artery was then embolized to stasis with 300-500 um Embospheres (Merit Medical, South Jordan, UT, USA). Post-embolization arteriogram confirmed stasis within the left bronchial artery and preserved patency of the superior phrenic artery branch. This resulted in resolution of the patient's hemoptysis for the duration of her hospitalization, and she was discharged home following completion of her course of intravenous antibiotics.

The patient re-presented to the emergency department 3 hours after discharge with recurrent massive hemoptysis. Inhaled tranexamic acid was administered, and the patient was admitted to the medical intensive care unit (MICU) with a plan for repeat embolization. A repeat CTA of the chest demonstrated an enlarging left upper lobe pseudoaneurysm with multiple systemic to pulmonary arterial fistulae (Fig. 1A-B). The patient was electively intubated, however diagnostic bronchoscopy was unable to definitively localize the source of bleeding. The patient then proceeded to pulmonary and systemic arterial angiography with embolization.

Via right common femoral vein access a pigtail catheter was advanced into the left main pulmonary artery and an angiogram showed normal patent branching of the left pulmonary arteries and no opacification of the pseudoaneurysm seen on the recent CTA (Fig. 2A).

Next, the right common femoral artery was accessed, and a catheter and guidewire combination was used to select the ostium of the left supreme intercostal artery. An angiogram demonstrated multiple tortuous abnormal vessels feeding a systemic to pulmonary arterial shunt (Fig. 2B) A microcatheter and wire were used to select a multiple distal left supreme intercostal artery branches, and embolization was performed with 500-700 um Embospheres. The microcatheter was removed, and a postembolization angiogram showed cessation of flow within the distal left supreme intercostal artery branch.

An attempt to recannulate the left supreme intercostal artery resulted in dissection and complete thrombosis of the left supreme intercostal artery as identified on post dissection angiogram.

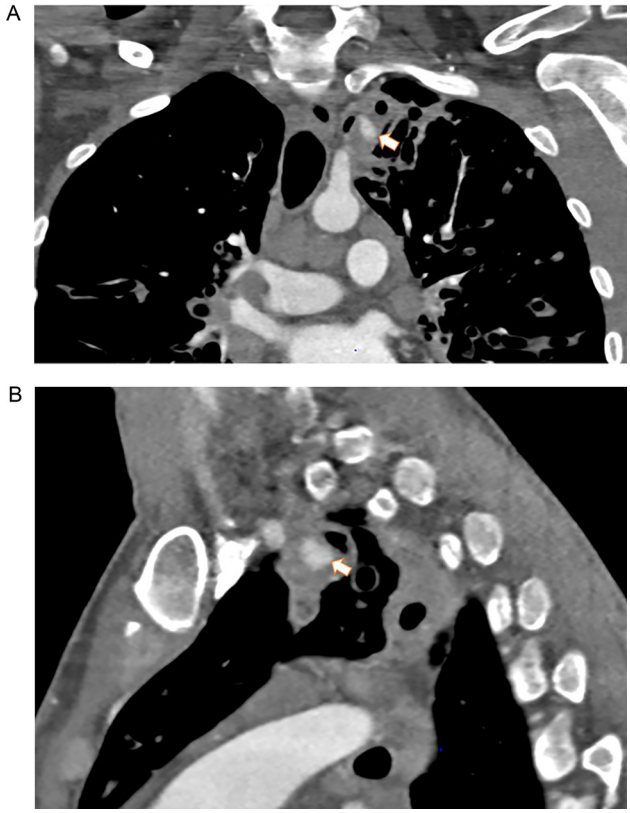

Fig. 1 – (A-B) CT angiogram of the chest at the time of initial presentation with hemoptysis demonstrating a pulmonary pseudoaneurysm (arrow) of unclear vascular supply seen in coronal (Fig. 1A), and sagittal planes (Fig. 1B).

The catheter was then used to select the left subclavian artery and left thyrocervical trunk. An angiogram showed a thyrocervical trunk with a large laterally directed distal branch supplying a systemic to pulmonary arterial fistula (Fig. 2C). An additional distal left thyrocervical trunk branch was seen to supply the pseudoaneurysm identified on the comparison CTA. The microcatheter and wire were advanced into the laterally directed distal branch of the left thyrocervical trunk and an angiogram showed numerous collaterals feeding the systemic to pulmonary arterial fistula. This was embolized to stasis with ONYX Liquid Embolic System (Medtronic, Minneapolis, MN, USA). A postembolization angiogram showed no flow across the systemic to pulmonary arterial fistula supplied by this lateral branch of the left thyrocervical trunk (Fig. 2D).

The microcatheter was withdrawn and was then used to select a superior medially directed branch of the left thyrocervical trunk. An angiogram from this microcatheter position showed a tortuous systemic to pulmonary arterial fistula supplying the pseudoaneurysm. Multiple attempts were made to cannulate the distal portion of this left thyrocervical trunk branch. This resulted in dissection and occlusion of this artery. A post dissection angiogram showed stasis within this superior medially directed branch of the left thyrocervical trunk.

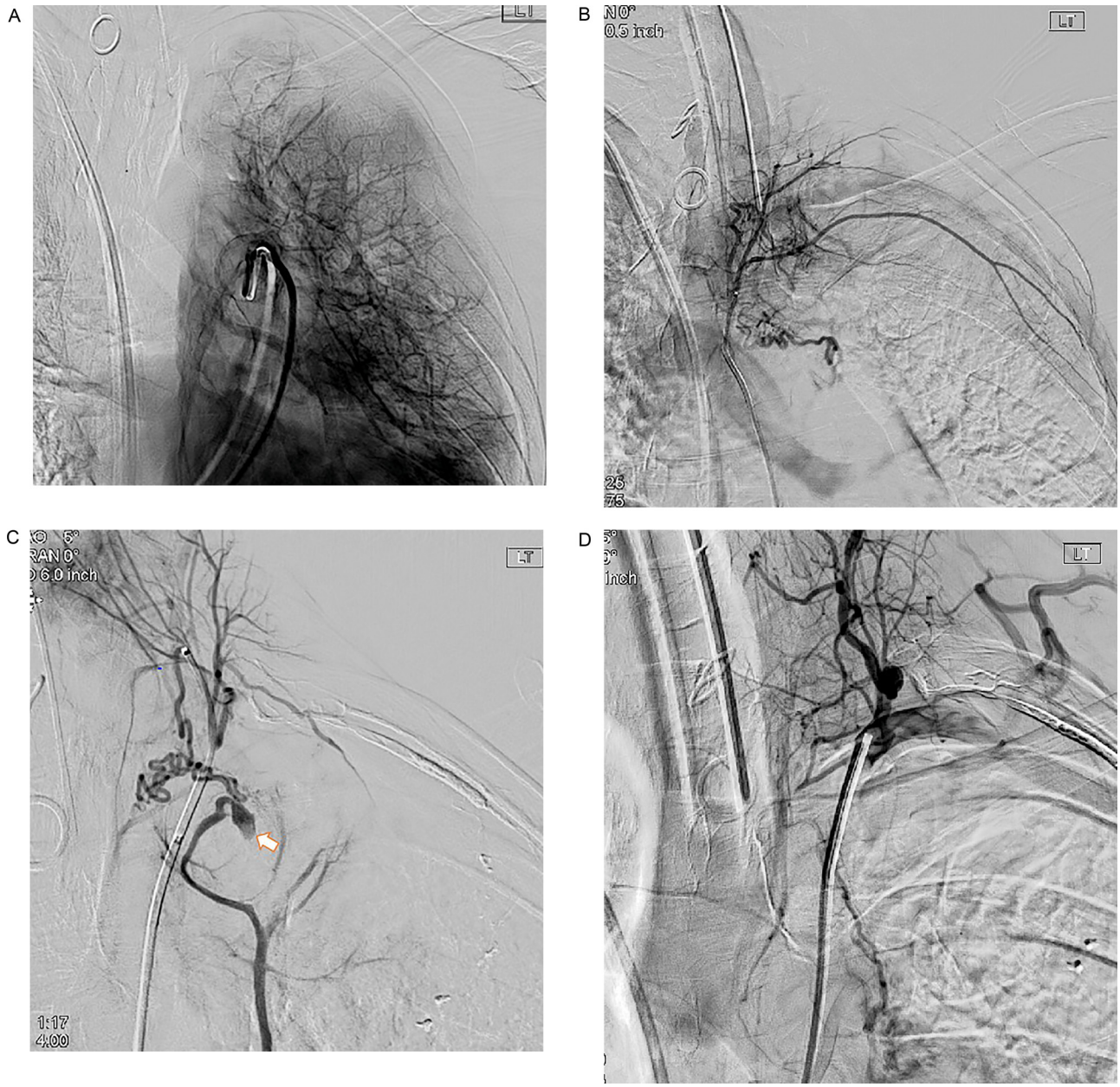

Fig. 2 – (A-C) Initial pulmonary artery angiography demonstrating no visible pulmonary artery aneurysm (Fig. 2A). After which, angiogram of the supreme intercostal artery showed tortuous systemic-to-pulmonary fistula (Fig. 2B), which was embolized along with several spinal arteries, but no aneurysm was seen. The thyrocervical artery was then selected, and angiography demonstrated system-pulmonary fistula with pseudoaneurysm (arrow) (Fig. 2C), which was then treated with particle embolization. Post-embolization angiography demonstrated lack of opacification of the pseudoaneurysm and fistula (Fig. 2D).

Through the femoral vein sheath, a 7 French angled pigtail catheter was then reinserted into the left main pulmonary artery and repeat left pulmonary arterial angiogram showed no newly visible pulmonary arterial supply to the pseudoaneurysm or persistent flow to the previously identified and embolized systemic to pulmonary arterial fistulae. The patient tolerated the procedure well and was transferred back to the MICU in stable condition.

The patient's hemoptysis improved, and she was extubated the following day. However, her clinical course was complicated by septic shock related to her CF exacerbation. Sputum cultures grew Burkholderia cepacian complex, pre-

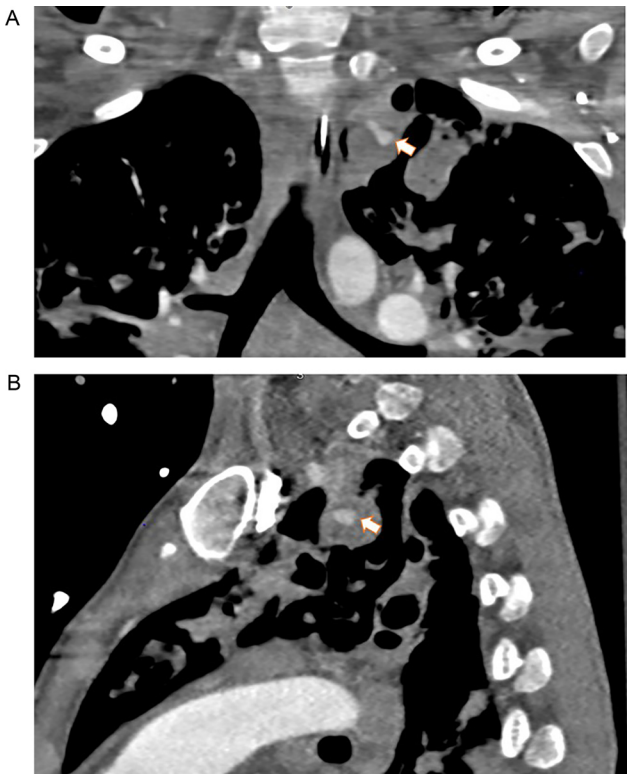

Fig. 3 – (A-B) Repeat CT angiogram of the chest at the time of re-presentation, 11 days after the original study. Coronal (Fig. 3A) and sagittal (Fig. 3B) sequences demonstrate grossly similar opacification the pseudoaneurysm (arrow), but now with increased soft tissue nodularity, and increased opacifications throughout the lung fields.

cluding lung transplantation. Eventually, she was weaned off vasopressors and transferred out of the MICU, remaining hemoptysis-free for 11 days. An additional isolated hemoptysis recurrence prompted transfer back to the MICU. A repeat CTA at that time did not show any active extravasation, although it did show continued enhancement of the pseudoaneurysm in the left lung apex (Fig. 3A-B). Following a goals of care discussion, the multi-disciplinary decision was made to actively monitor, while having a low threshold to proceed with additional emergency embolization if the patient decompensated versus elective embolization. One week later the patient developed recurrent massive hemoptysis requiring intubation. Repeat CTA redemonstrated the 8-9 mm pseudoaneurysm in the medial left upper lobe without evidence of active extravasation, but with altered morphology compared to the most recent prior CTA. The decision was made to proceed with repeat embolization.

Via right common femoral vein access a 7 French angled pigtail catheter was inserted and advanced through the right side of the heart into the left pulmonary artery. From this catheter position, angiography revealed a patent and unremarkable left pulmonary artery. The catheter was then advanced into the pulmonary artery branch supplying the left upper lobe. Angiography from this catheter position revealed opacification of the pseudoaneurysm seen on recent CTA.

A microcatheter was then advanced over a wire into a distal branch of the pulmonary artery supplying the left upper lobe. From this microcatheter position, an angiogram was performed, confirming opacification of the pseudoaneurysm (Fig. 4A).

Using a Synchro-2 microwire and Excelsior SL 10 microcatheter combination (Stryker Corporation, Kalamazoo, MI, USA), the microcatheter was advanced through the medial distal branch of the left upper lobe pulmonary artery (Fig. 4B), through the pseudoaneurysm, and into the systemic arterial inflow to the pseudoaneurysm supplied by the previously dissected left thyrocervical trunk arterial branch (Fig. 3C). Angiography from this catheter position, confirmed opacification of a patent collateral arteries originating from the left thyrocervical trunk. Coil embolization of this systemic arterial feeding artery, the pseudoaneurysm itself, as well as the distal pulmonary artery branch was performed (Fig. 3D-E). A post embolization angiogram revealed stasis within the PAP and its feeding arteries (Fig. 3F). The patient tolerated the procedure well, without any immediate post procedure complications.

Although the procedure was successful technically, the patient continued to deteriorate clinically, requiring increased respiratory support with maximal ventilator settings and pressor support. On post-procedure day four, the family made the decision to transition to comfort measures only due to poor prognosis. The patient expired shortly thereafter with the cause of death being acute respiratory failure due to Burkholderia cepacian infection, a complication of her cystic fibrosis.

Discussion

PAP vascular supply can be highly variable including pulmonary arterial, bronchial arterial, and non-bronchial systemic arterial supply. Consequently, the standard approach to endovascular treatment involves embolization of both the pulmonary and systemic feeding vessels of the PAP. Additionally, this variability in vascular supply highlights the value of CTA evaluation in patients suspected of having PAP, as it allows for early identification of the pseudoaneurysm location, origin of the bronchial artery, and identification of any non-bronchial artery systemic supply to the PAP [7].

Shin et al classified PAPs into four angiographic sub-types [6]. PAPs identified by non-selective pulmonary angiography were defined as type A. Type B PAPs were defined as pseudoaneurysms identified via selective segmental or sub-segmental pulmonary angiography. PAPs identified by bronchial and non-bronchial arterial angiography due to bronchopulmonary shunt and not identified on pulmonary angiography were classified as type C. Type D PAPs were defined as PAPs identified on CTA, but not visible on conventional angiography.

This case demonstrates the challenge of treating type C PAPs due to the lack of visualization on pulmonary arterial angiography. In this case, the augmentation of systemic arterial supply by prior embolization resulted in transformation of the type C PAP into a type B PAP. This facilitated visualization of the PAP on repeat pulmonary arterial angiography, an ultimately allowed for advancement of the microcatheter across

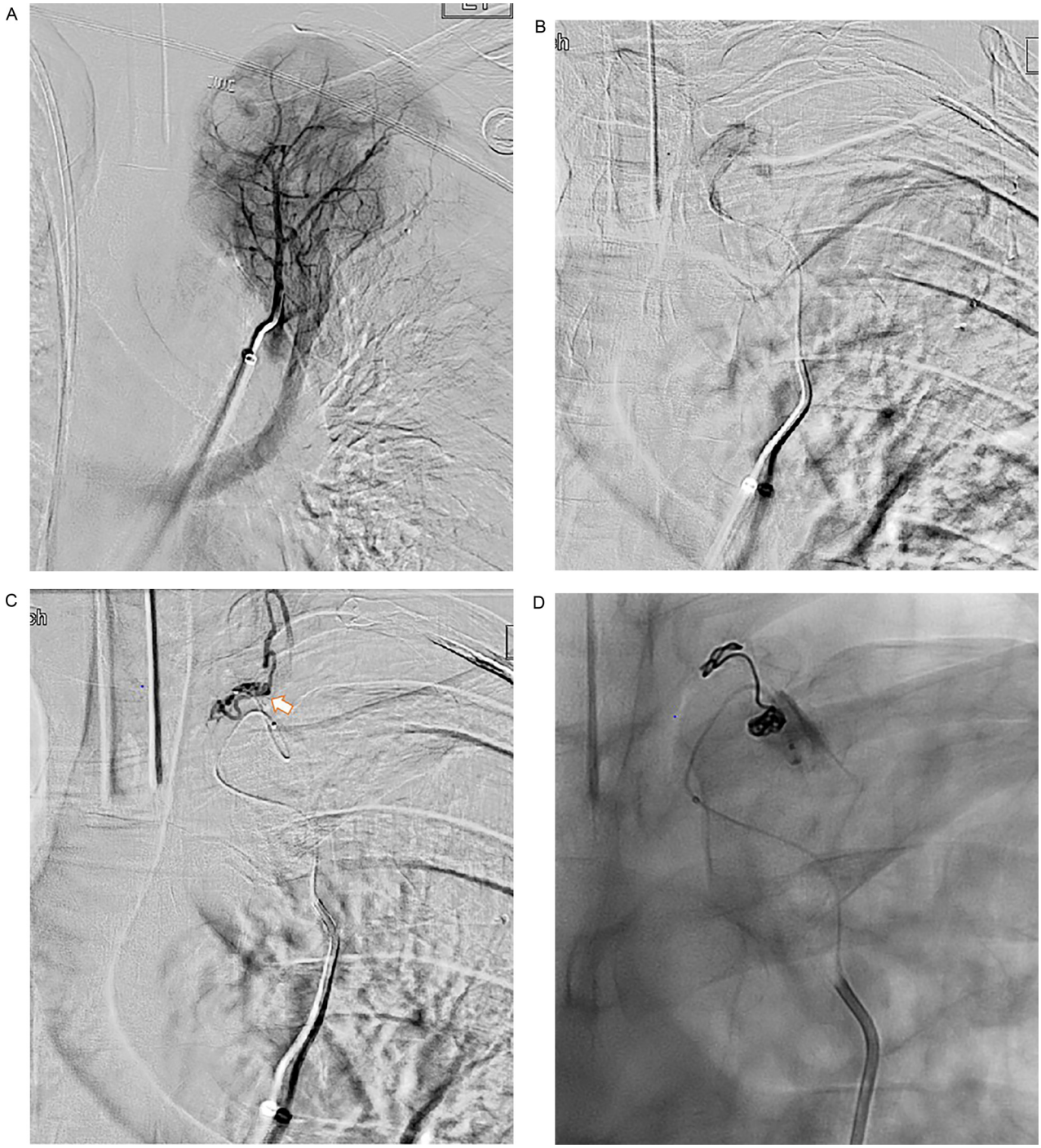

Fig. 4 – (A-F) Angiography of the pulmonary artery 11 days later then demonstrates the aneurysm in the left apex(Fig 4A). Through a 7F sheath, a Synchro 2 wire and an SL 10 microcatheter (Stryker Corporation, Kalamazoo, MI, USA) were advanced into the medial distal branch of the left upper lobe pulmonary artery (Fig. 4B). The microcatheter was then advanced through the pseudoaneurysm, and into the systemic arterial inflow of the feeding thyrocervical artery (arrow) (Fig. 4C). Coil embolization of branches of the thyrocervical trunk and the pseudoaneurysm was performed (Fig. 4D), followed by embolization of the feeding branch of the pulmonary artery (Fig. 4E). Final angiogram demonstrated resolved opacification of the pulmonary artery pseudoaneurysm (Fig. 4F).

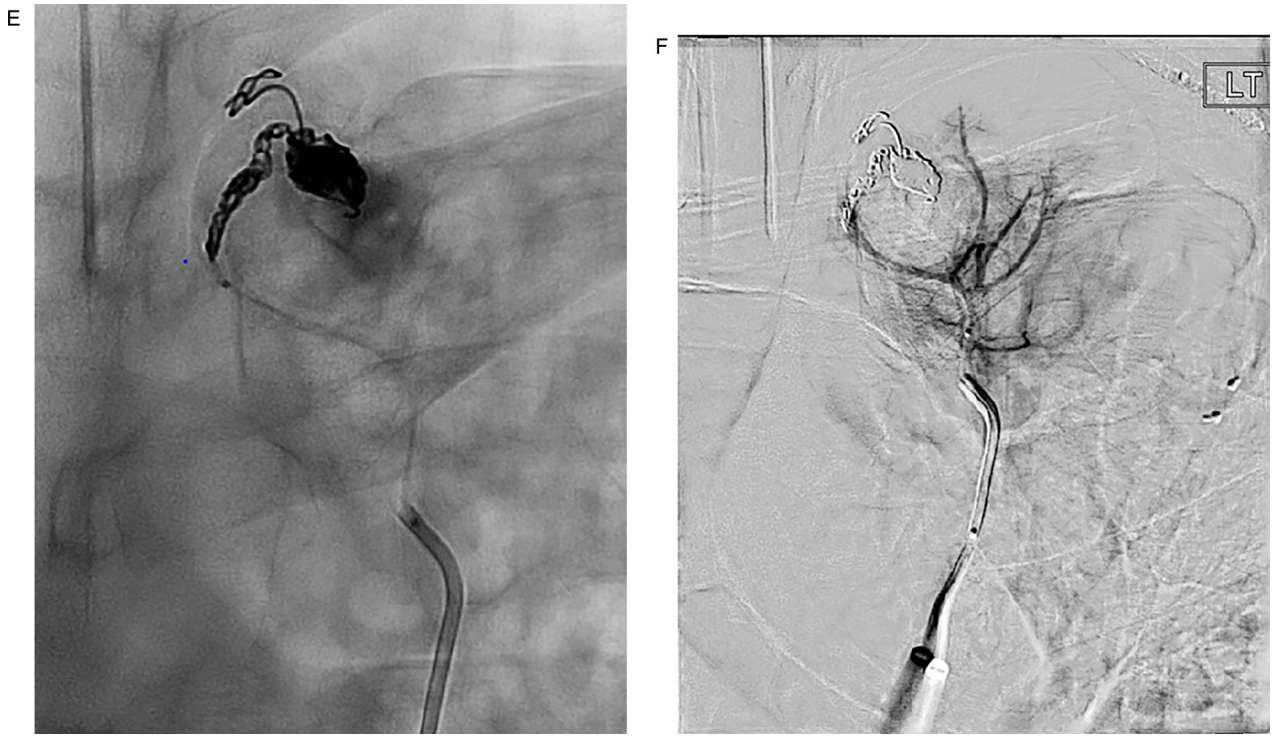

Fig. 4 – Continued

the pseudoaneurysm and into the system arterial inflow. Furthermore, in their case series of six PAPs, Tsukada et al discussed inflow augmentation and flow reversal following systemic arterial embolization and made recommendations for embolization strategies based off PAP sub-type [8]. Our experience in the case report validates their recommendations.

Additionally, we support efforts to further understand the unique characteristics of PAPs based off their angiographic classification system and what additional endovascular interventions should be taken to assure adequate hemostasis in PAPs with unusual vascular supply. Future reports and case studies should attempt to categorize PAPs based off the established classification system (Shin et al) [6], as this would allow for easier comparison of outcomes between endovascular interventions across PAP subtypes.

Case concept

GS; case/literature review, manuscript drafting and revision: all authors

Ethical approval

All procedures performed in the studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Patient Consent

Patient consent was obtained. There are no identifiable images in the report. All procedures performed in the studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

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