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Case Report

Successful embolization of an infected aneurysm of a subsegmental pulmonary artery in an infant with necrotizing MRSA pneumonia

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ARTICLE INFO

Article history: Received 13 April 2019 Revised 8 June 2019 Accepted 17 June 2019 Available online 4 July 2019

Keywords: Pulmonary artery aneurysm Pneumonia Microvascular plug

ABSTRACT

Para-infections aneurysms are a very rare complication but bear the risk of significant morbidity and mortality in case of rupture and hemorrhage. We present the youngest published case of a right-sided pulmonary artery pseudoaneurysm due to nonmultiresistant *Staphylococcus aureus* pneumonia in a 7-month old boy, complicated by 2 episodes of significant hemorrhage. Selective microvascular plug embolization of the feeding segmental pulmonary artery by interventional radiology and cardiology was successfully undertaken while having a cardiothoracic surgical team on stand-by. Follow-up ultrasounds showed no residual flow distal to the microvascular plug. The patient had complete clinical recovery 10 months after the initial presentation. Interventional radiology procedures are challenging in children due to limited availability of appropriately-sized equipment, low case numbers, and a limited body of literature.

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Work was performed at the Paediatric Intensive Care Unit, Queensland Children's Hospital, 501 Stanley Street, South Brisbane, QLD, Australia.

Conflict of Interest: The authors declare that they have no conflict of interest.

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https://doi.org/10.1016/j.radcr.2019.06.005

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Introduction

Acquired pulmonary artery aneurysms (PAA) are a very rare entity. The incidence remains unclear in children. The youngest case documented at necropsy was by Zuber in 1904 involving a 5-month-old child who had developed multiple parapneumonic aneurysms of the pulmonary trunk, the main pulmonary arteries as well as smaller branch arteries [1].

PAAs can be idiopathic or caused by infection (tuberculous and nontuberculous), trauma (extra- and endovascular), neoplasms, postballoon angioplasty (with or without stent), Swan-Ganz-Catheter, angiography, pulmonary artery banding, chest tube insertion or lung biopsy, systemic vasculitis (especially Behçets disease), atherosclerosis, pulmonary hypertension and collagen and connective tissue disorders such as Marfan's disease or Williams-Beuren syndrome [2–4].

The most commonly isolated organisms include Streptococcus and Staphylococcus species. Fungal pathogens include Aspergillus, Candida and Mucor and are overall very rare but more common in patients with congenital heart defects, diabetes mellitus, and hematological malignancies [5].

Pulmonary arteries can be classified into central and peripheral. Central PAs include the pulmonary trunk as well as the main left and right PAs. The intraparenchymal arteries are classified as peripheral PAs. Aneurysms of the latter are considered a life-threatening condition and warrant immediate investigation and treatment. Laplace's law states that wall stress and therefore risk of rupture increases with pressure and vessel radius. Greater vessel wall thickness is thus a protective factor. Mechanisms contributing to the formation of pulmonary artery aneurysms in lung infections include embolization of the vasa vasorum, blockage of vascular lumina by septic emboli, and erosion of the vessel wall by inflammatory processes. Differentiation between true aneurysms involving all 3 vessel wall layers and pseudoaneurysms can only be made histologically.

Hemoptysis or pulmonary hemorrhage are the most common presenting symptoms and warrant immediate diagnostics and treatment. Other clinical features include a systolic murmur, fever in case of infection, dyspnea, and cough. Gold standard for diagnosis is spiral CT with pulmonary angiography and ability for 3D reconstruction followed by percutaneous pulmonary arteriography. The classic appearance of PA aneurysms is rapid filling with delayed emptying.

Case report

A 7-month-old boy from the Torres Strait Islands with no significant past medical history presented to the local clinic with a 3-day history of coryzal symptoms, cough, fever, and poor feeding. A chest x-ray showed a large right-sided pleural effusion with mediastinal shift to the left (Fig. 1). He was retrieved to the Paediatric Intensive Care Unit (PICU) at the closest tertiary hospital, where a chest ultrasound showed a large rightsided effusion, approximately 3 cm deep with a few septations but no loculations. He was intubated to facilitate procedures and had a right-sided intercostal pigtail catheter and internal

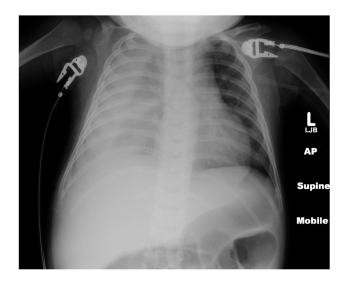


Fig. 1 – CXR on presentation showing a large right-sided pleural effusion with mediastinal shift to the left.

jugular central venous line inserted. The drain yielded 180 mL of sero-purulent fluid with Gram-positive cocci on microscopy. The patient received twice daily urokinase dwells for 3 days and was extubated the next day, initially to high flow and subsequently to low flow nasal oxygen.

Five days later, a right sided pneumothorax occurred despite the chest drain being in situ. An additional, anterior chest drain was inserted, with resolution of the pneumothorax. The pleural fluid microbiology revealed nonmultiresistant Methicillin-resistant Staphylococcus aureus. Antimicrobial treatment initially consisted of Cefotaxime and Vancomycin which was subsequently changed to oral Cotrimoxazole due to clinical improvement, difficulty maintaining therapeutic Vancomycin levels and consistency with sensitivities.

Ten days after the initial presentation, there was acute pulmonary hemorrhage with an estimated 130 mL of frank blood draining from his chest drain. He required emergent intubation and high ventilatory pressures as well as a packed red blood cell transfusion which achieved stabilisation. CT pulmonary angiogram showed a large peripulmonary hematoma and aneurysmal dilatation (approximately $1.8 \times 4.1 \times 10.5$ cm) of the subsegmental branch of the artery supplying the right lower lobe (Fig. 2). He had a further hemorrhage the following morning during spontaneous coughing and was therefore re-paralysed with good effect and no further pulmonary bleeding. He was started on tranexamic acid and his chest drain was converted to a larger 16 Fr intercostal catheter due to blockage of the pigtail with a clot.

He was then retrieved to the PICU at a quaternary centre for further intervention. A repeat CT pulmonary angiogram showed interval growth of the pseudoaneurysm and significant necrotic areas of the right lower lung (Fig. 3). Given the precarious situation and his relative stability with no further significant bleeding, he was monitored with daily chest ultrasounds to assess for potential interval growth of the lesion (Fig. 4). There were ongoing multidisciplinary discus-



Fig. 2 – Initial CT angiogram showing aneurysmal dilatation of right lower lobe segmental pulmonary artery with adjacent inflammatory changes of the pulmonary parenchyma.

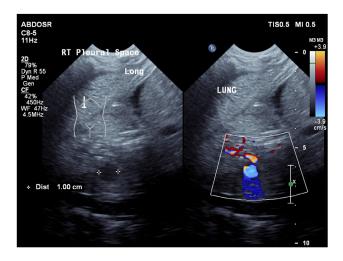


Fig. 4 – Ultrasound of chest showing pseudoaneurysm of right lower lobe segmental pulmonary artery with internal flow on color Doppler imaging.

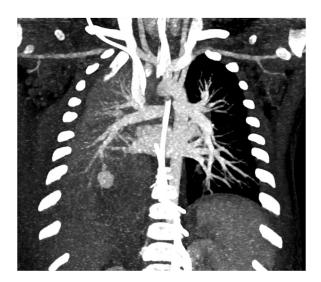


Fig. 3 – Repeat CT angiography showing interval growth of R lower lobe pseudoaneurysm.

sions between interventional radiologists, general surgeons, cardiothoracic surgeons, and respiratory physicians about the best approach, with some of the concerns including the risk of pulmonary artery rupture, need for lobectomy due to necrosis and to goal to avoid pneumonectomy. He subsequently underwent a combined interventional cardiology and interventional radiology approach with general surgical and cardiothoracic surgical teams on standby. A left groin common femoral venous puncture was performed followed by placement of a 5F hemostatic sheath. Right ventricular outflow tract and main pulmonary artery were catheterized with a 5F Lehman ventriculography catheter (Medtronics. A pseudoaneurysm in the

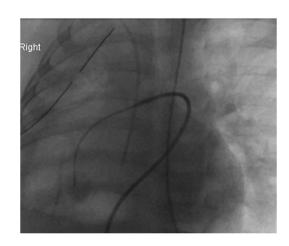


Fig. 5 – Pseudoaneurysm in the posterior segmental branch of right lower lobe during angiography.

posterior segmental branch of right lower lobe was identified on angiography (Fig. 5). The segmental feeding vessel was then selectively catheterized with a Progreat microcatheter (Terumo) and embolized with a microvascular plug (MVP3, Medtronic) (Fig. 6). Final angiography showed no residual flow in the pseudoaneurysm but preserved flow in the remainder of the segmental branches of the right lower pulmonary artery (Fig. 7).

Follow-Up ultrasound in PICU showed no residual flow within the aneurysm (Fig. 8). His chest drain was removed the following day, 2 days later he was extubated and 10 days later he was discharged home. He completed a 6-week course of oral Co-Trimoxazole. Ten months following his initial presentation he was thriving without ongoing respiratory symptoms or recurrent infections. Besides some residual pleural thickening, there was no evidence of sequelae on the follow-up chest x-ray (Fig. 9). Immune function tests showed normal results for lymphocyte subsets and neutrophil function.

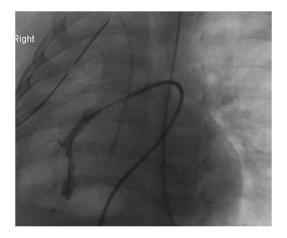


Fig. 6 – Selective catheterization of the segmental feeding vessel with a Progreat microcatheter (Terumo) and embolization with a microvascular plug (MVP3, Medtronic).

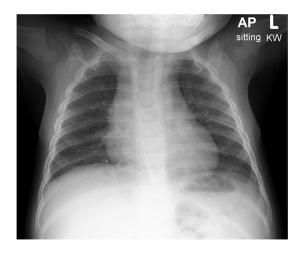


Fig. 9 – Follow-up chest x-ray 11 months after the initial presentation showing only mild residual pleural thickening without evidence of other sequelae.

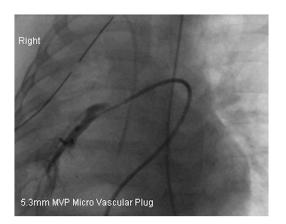


Fig. 7 – Final angiography showing no residual flow in the pseudoaneurysm but preserved flow in the remainder of the segmental branches of the right lower pulmonary artery.

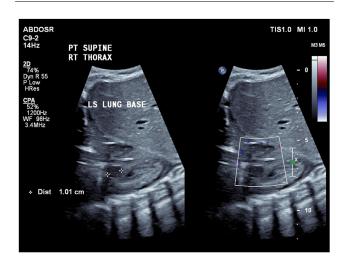


Fig. 8 – Follow-up ultrasound showing complete thrombosis of the pseudoaneurysm sac with no residual flow.

Discussion

Traditionally peripheral PAAs have been treated with lobectomy or aneurysmectomy. Currently, the endovascular approach with coil embolization is preferred as it is less invasive and associated with fewer complications in adults. Amplatzer devices have been successfully used to treat peripheral PAAs as well as arteriovenous malformations in adults and children [6–8]. In contrast, surgical intervention remains the main therapeutic approach to lesions of the main pulmonary artery.

To our knowledge this case describes the youngest reported patient undergoing successful endovascular embolization of a subsegmental pulmonary artery in the setting of a parainfectious aneurysm. This extremely rare complication should be considered in any patient with pneumonia and acute onset of pulmonary hemorrhage of unknown origin. CT angiography with 3-D reconstruction facilitates definition of the anatomy and allows interventional planning.

Selective endovascular occlusion of the feeding artery can be achieved but consideration should be given to secondary options to cover for procedural complications and/or inability to occlude the vessel. Close collaboration between interventional cardiology, interventional radiology, and cardiothoracic surgery is pivotal to optimize patient outcomes. Interventional radiology procedures are challenging in children because most of the equipment is designed for adults. Catheters are often longer than the height of a child and too big for their vessels. Fluid balance and contrast doses are particularly important in children. Careful measurement of contrast injections and other fluid infusions (on a mL/kg basis) is crucial to avoid fluid overload in small children. Optimization of imaging techniques is necessary to reduce the radiation dose in this vulnerable patient group.

Informed consent

Informed consent was obtained from all individual participants in the study.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.radcr.2019.06.005.

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