Transcatheter Embolization of Systemic-to-pulmonary Artery Fistulas: a Case Report

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Case report

Keywords: systemic artery to pulmonary vessel fistula (SAPVF), Transcatheter embolization, abnormal communication, case report

Posted Date: November 8th, 2021

DOI: https://doi.org/10.21203/rs.3.rs-1004874/v1

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Abstract

The systemic artery to pulmonary vessel fistula (SAPVF) is an uncommon vascular abnormal communication between systemic arteries (except bronchial arteries) and the lung parenchyma[1]. It can be divided into congenital and acquired causes. Congenital SAPVF is often accompanied by cardiac or pulmonary artery hypoplasia, and acquired are usually caused by pleural adhesions after pleurisy, empyema, trauma, or surgery[2].

We report a case of transcatheter arterial embolization for the treatment of congenital right inferior phrenic artery to pulmonary artery fistula.

Case Presentation

A 32-year-old man was admitted to the hospital with recurrent hemoptysis. He was previously healthy and did not have any family history of hemoptysis. He had repeated hemoptysis without obvious causes for more than a week and condition aggravation in the morning and evening, but no cough, expectoration, fever, nor dyspnea. Upon examination, there was no obvious abnormality. A preoperative nonenhanced computed tomography (CT) scan of the chest showed that a soft tissue density stripe abutting on the oblique fissure in the left lower lobe (Figure 1A). And the contrast-enhanced CT scan revealed about a 1.3 cm-sized well-enhanced vascular mass and an abnormally connection between the thickened right inferior phrenic artery and the branch of the pulmonary artery of the left lower lobe(Figure 1B). A systemic (right inferior phrenic artery)-pulmonary artery (branch of the pulmonary artery in the anteromedial basal segment of the left lower lobe) fistula was diagnosed.

The patient refused the operation and had received embolization treatment. Prior to embolization, vascular malformation was confirmed by selective angiography. The views have tortuous and thickened right inferior phrenic artery(Figure 1C) and malformed anastomosis at the end of left inferior pulmonary artery display(Figure 1D) but without obvious pulmonary vein (Figure 2A). During the diagnostic angiography, 1.5 ml of iodized oil and 0.5 ml of tissue glue embolization were performed with a Superselective microcatheter. After embolism, there was no blood flow or a little blood flow(Figure 2B) and soon the patient's symptom disappeared. No vascular abnormality was visible three and one half months later when a plain and enhanced CT were done and the patient had no particular symptoms.

Discussion

SAPVF can be divided into congenital SAPVF and acquired SAPVF. About 50% of SAPVF are congenital and are more common in patients with cardiopathy or pulmonary artery hypoplasia[1]. Acquired SAPVF is mostly caused by tumors of the pleura or lung, inflammation, or iatrogenic chest injury, which obstructs the downstream obstruction of the involved vessels and creates collateral circulation in the chest wall or mediastinum.
These vascular fistulas are usually supplied by the internal thoracic and intercostal arteries, but also from the internal mammary and abnormal aortic branches, subclavian, axillary, phrenic, mediastinal, and coronary arteries\textsuperscript{[3, 4]}. Afferent arteries can be single or multiple and multiple systemic arteries to pulmonary artery fistulas without any other underlying diseases are extremely rare\textsuperscript{[5]}.

Most patients had no particular symptoms and the clinical features were continuous murmurs on chest auscultation or abnormal X-ray findings found in the patients in physical examination. Serious cases may have breathlessness, congestive heart failure, or hemoptysis\textsuperscript{[1]}. When the systemic branch is the coronary artery, symptoms such as chest pain, exertional dyspnea, palpitations, angina pectoris, and acute inferior myocardial infarction may occur\textsuperscript{[6]}. And if there are multiple systemic to pulmonary artery fistulas, there could be severe pulmonary hypertension\textsuperscript{[7]}.

Selective angiography is the reference standard for diagnosing SAPVF. It can make the complete appearance of the anatomic structure of the lesion more obvious and depict the extent and location of lung involvement. The contrast-enhanced CT also plays an important role in finding the causes of SAPVF and SAPVF related complications; therefore, it should be routinely reserved for SAPVF.

Treatment indications remain debated because the natural history of SAPVF is not well known. In patients who are unexpectedly found to have asymptomatic SAPVF, some authors believe that intervention is necessary because of the risk of bacterial infection, bleeding (dilation and rupture), and heart disease (pulmonary hypertension or congestive heart failure)\textsuperscript{[4, 5]}.

The main treatment methods of SAPVF are including embolization, surgical treatment, or observation. Recently, embolization has become the preferred method because it causes minor trauma, minimal loss of lung parenchyma, and does not require general anesthesia\textsuperscript{[5]}. It was reported that when SAPVF has few inflow arteries, surgical treatment has a better effect than embolization. However, surgical is controversial in SAPVF with multiple arterial infusions, because of the high risk for bleeding complications\textsuperscript{[1]}. And comprehensive surgical treatment should be fully evaluated, particularly patients combined with other complex cardiovascular diseases\textsuperscript{[8]}.

Conclusions

In conclusion, we described our experience with a case of transcatheter arterial embolization for the treatment of congenital SAPVF. This treatment is a safe, less invasive, and a reasonable therapeutic option to control bleeding complications.

Declarations

Ethics approval and consent to participate: Written informed consent was obtained from the individual(s) for the publication of any potentially identifiable images or data included in this article.
Availability of data and materials: The raw data supporting the conclusions of this article will be made available by the authors, without undue reservation.

Competing interests: The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

Funding: This study was not supported by any funding.

Authors’ contributions: All authors listed have made a substantial, direct and intellectual contribution to the work, and approved it for publication.

Acknowledgements: None.

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Figures

Figure 1

(A) CT pulmonary window showed a soft tissue density strip abutting on the oblique fissure in the left lower lobe. (B) A contrast-enhanced CT scan showed a malformed vascular mass and significantly thickened right phrenic artery. (C) Selective angiography of the right inferior phrenic artery. (D) Selective angiography of the left pulmonary artery.
Figure 2

(A) Angiogram of the right phrenic artery and the left pulmonary artery. (B) After embolism, there was no blood flow or a little blood flow.