



## Case report

# Successful transcatheter arterial embolization of asymptomatic aneurysm associated with left inferior phrenic artery-to-left pulmonary artery fistula: A case report

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## ABSTRACT

Cases of inferior phrenic artery-to-pulmonary artery fistulas and those complicated by massive hemoptysis have been rarely reported. A 38-year-old man presented to our hospital with a chief complaint of coughing. Computed tomography (CT) revealed a nodule in the left lower lobe, and contrast-enhanced CT showed inflow of contrast medium into the nodule. CT angiography detected an aneurysm associated with a left inferior phrenic artery-to-left pulmonary artery fistula. Transcatheter arterial embolization (TAE) was performed to prevent hemoptysis. Hemoptysis did not occur during the 2-year follow-up. We report a rare case of asymptomatic aneurysm associated with a left inferior phrenic artery-to-left pulmonary artery fistula, which was successfully treated using TAE to prevent hemoptysis.

## 1. Introduction

Inferior phrenic artery-to-pulmonary artery fistulas are rare, and few reports have identified it as a cause of major hemoptysis [1,2]. Transcatheter arterial embolization (TAE) is a well-established, minimally invasive therapeutic modality for the management of hemoptysis [3]. Herein, we report a rare case in which an asymptomatic aneurysm associated with a left inferior phrenic artery-to-left pulmonary artery fistula was successfully treated using TAE to prevent hemoptysis.

## 2. Case presentation

A 38-year-old man presented to our hospital with a chief complaint of coughing for a few days. The patient had no history of illness, trauma, or smoking. Chest X-ray screening yielded normal findings; however, computed tomography (CT) revealed a nodule in the left lower lobe (Fig. 1A). Contrast-enhanced CT showed an inflow of contrast medium into the nodule as well as a connection between the nodules and pulmonary artery (Fig. 1B and C). CT angiography detected an aneurysm

associated with a left inferior phrenic artery-to-left pulmonary artery fistula (Fig. 1D). The cough disappeared over the course of investigation.

TAE was performed to prevent hemoptysis. Although the aneurysm was not imaged from the left pulmonary artery (Fig. 2A), it was found to be associated with the left inferior phrenic artery-to-left pulmonary artery fistula when observed in the left phrenic artery (Fig. 2B). We performed coil-assisted embolization of the inferior phrenic artery using n-butyl cyanoacrylate-lipiodol, after which angiographic examination of the left inferior phrenic artery no longer showed inflow of contrast medium into the aneurysm (Fig. 2C).

During follow-up a month after the TAE was performed, a chest X-ray examination indicated that the location of the embolic material was not of major concern (Fig. 3). Over the course of 2 years of follow-up, hemoptysis did not occur.

## 3. Discussion

In this report, we present a rare case in which an asymptomatic aneurysm associated with a left inferior phrenic artery-to-left pulmonary

*Abbreviation:* TAE, Transcatheter arterial embolization; CT, computed tomography.

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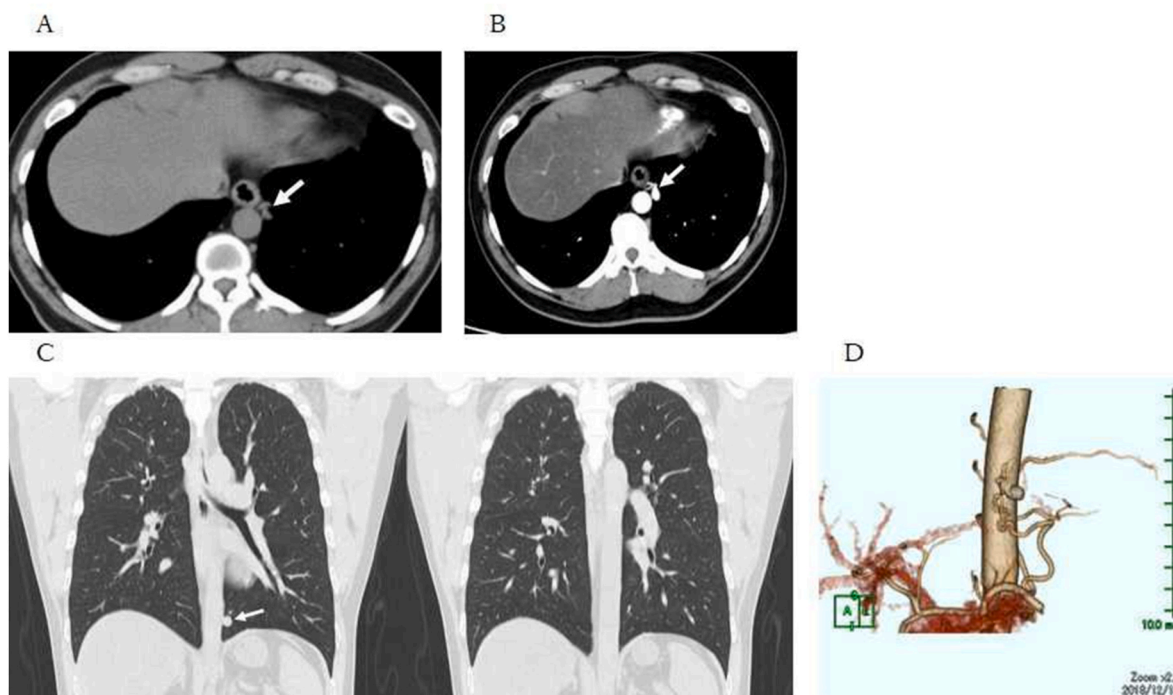
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**Fig. 1.** (A) Computed tomography image showing a nodule in the left lower lobe. (B) Contrast-enhanced computed tomography showing inflow of contrast medium into the nodule and (C) the connection between the nodules and pulmonary artery. (D) Computed tomography angiography showing the aneurysm associated with the left inferior phrenic artery-to-left pulmonary artery fistula.



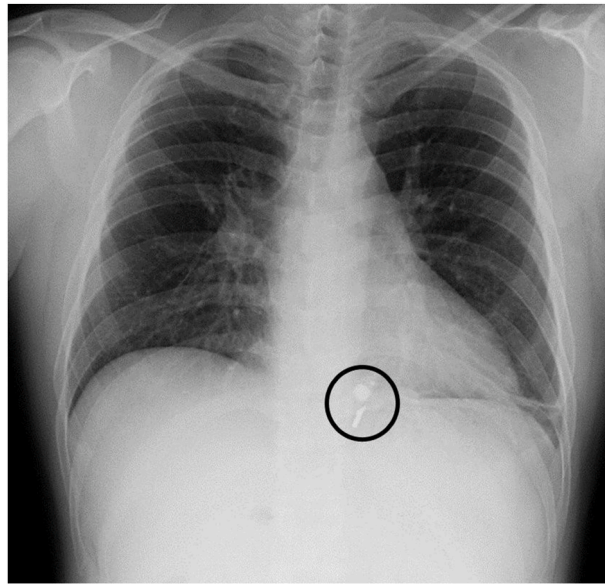
**Fig. 2.** (A) Angiography of the left pulmonary artery did not indicate the presence of an aneurysm. (B) Angiography of the left inferior phrenic artery revealed the presence of an aneurysm associated with a left inferior phrenic artery-to-left pulmonary artery fistula. (C) Following embolization, there was no inflow into the aneurysm.

artery fistula was successfully treated using TAE to prevent hemoptysis. At the first visit the patient had cough; however, it disappeared during the scrutiny process. Therefore, in this case, we considered the symptoms and illness to be unrelated.

Abnormal systemic-to-pulmonary shunts mainly develop due to chronic inflammation, trauma, hypoxic vasoconstriction, or arterial thrombosis [2,4]. Notably, our patient had no history of illness or smoking; therefore, it is conceivable that the patient's condition was congenital.

We performed a search of the PubMed database using the following terms: "phrenic artery," "pulmonary artery," and "fistula." Our search yielded nine articles. After excluding two articles that were not written in English and those that did not specifically relate to inferior phrenic artery-to-left pulmonary artery fistulas, we reviewed four articles discussing a total of five cases (Table 1) [1,2,5,6].

Among these five cases, the median age was 52 years (range, 38–82 years). Two cases involved men, whereas the other three involved women. Massive hemoptysis was reported in two cases, both of which showed improvements following TAE. In three of the cases, the fistulas were located on the right side; in the remaining two cases, they were on the left side. Notably, ours was the only case that was complicated by an aneurysm. To the best of our knowledge, this is the first report to discuss an asymptomatic aneurysm associated with a left inferior phrenic artery-to-left pulmonary artery fistula. To prevent the aneurysm from leading to massive hemoptysis, we performed TAE, as we believed this option to be less invasive than other surgical measures. We considered that performing prophylactic TAE for aneurysm associated with inferior phrenic artery-to-pulmonary artery was important because this was a shunt from the aorta and had a risk of massive hemoptysis and critical clinical course. In this case, TAE carried a risk of pulmonary embolism and



**Fig. 3.** During follow-up, a month after the treatment, a chest X-ray revealed the presence of embolic material.

**Table 1**

Literature review of reports on inferior phrenic artery-to-pulmonary artery fistulas and their clinical findings.

No.	Year	Authors	Country	Age (years)	Sex	Symptom	Side	Aneurysm	Treatment	Outcome
1	2011	Shao-Jung Hsu et al.	China	42	Male	Massive hemoptysis	Left	–	TAE	Improved
2	2017	Emi Yakushiji et al.	Japan	82	Female	Massive hemoptysis	Right	–	TAE	Improved
3	2019	Arthur Soares Souza Jr et al.	Brazil	52	Female	Asymptomatic	Right	–	ND	ND
4	2019	David Livingston et al.	USA	59	Female	Dyspnea	Right	–	Watch	ND
5	2021	Current case	Japan	38	Male	Asymptomatic	Left	+	TAE	Improved

TAE: transcatheter arterial embolization, ND: not described.

penetration during treatment. Hence, preparing for emergency surgery in case of side effects and application choice was important.

#### 4. Conclusion

We reported a case in which an asymptomatic aneurysm associated with a left inferior phrenic artery-to-left pulmonary artery fistula was treated using transcatheter arterial embolization, and in this case, we considered that performing prophylactic TAE for aneurysm associated with inferior phrenic artery-to-pulmonary artery was important.

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#### Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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