Left Pulmonary Vein Thrombosis after Left Lower Lobectomy in a Case of Adulthood Type 2 Congenital Cystic Adenomatoid Malformation Presenting with Left Hemiparesis

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Abstract

Pulmonary vein thrombosis (PVT) is a known complication after lung transplantation but has rarely been reported after lower lobectomy or bilobectomy. We report a case of a left PVT extending into left atrium along with left hemiparesis following an uneventful left lower lobectomy in a case of adult congenital cystic adenomatoid malformation type 2 (CCAM). CCAM being rare in adults further its association with PVT following lower lobectomy makes this case an unusual interesting case.

Keywords
- left pulmonary vein thrombosis
- adult congenital cystic adenomatoid malformation type 2
- lower lobectomy

Introduction

Pulmonary vein thrombosis (PVT) is rarely reported after lower lobectomy.[1] Adult congenital cystic adenomatoid malformation Type 2 (CCAM) is rare in adults.[2] Here we present an unusual and interesting case of left lower lobectomy that was performed due to CCAM, which further led to the development of PVT and hemiparesis.

Case Description

A 40-year-old female presented with complaints of fever of 6 months duration, which was high grade and continuous, associated with cough, expectoration, and chest pain. Initial chest X-ray (►**Fig. 1**) was suggestive of left pleural effusion, for which patient underwent a pleural tapping and was symptomatically better after the initiation of therapy. After 10 days, patient started to have fever again and underwent contrast enhanced computerized tomography (CECT) thorax (►**Figs. 2** and **3**) which was suggestive of a well-defined, hypodense, and thin-walled nonenhancing uniloculated lesion-posterior basal segment of left lower lobe indicative of intralobar sequestration, having arterial supply to the lesion by two small branches arising from descending thoracic aorta and venous drainage into the pulmonary circulation (►**Fig. 4**). Bronchoscopy was done to rule out endobronchial communication of the lesion that showed no communication, thus indicating intralobar sequestration (►**Fig. 5**). Hence, patient underwent left lower lobectomy which was uneventful intraoperatively and postoperatively. The patient was discharged on postoperative day 6.

The tissue was subjected for histological examination, and diagnosis of congenital cystic adenomatoid malformation-type 2 was made (►**Figs. 6** and **7**).

In total, 4 days of postdischarge, the patient again presented in the casualty with complaints of left hemiparesis (upper and lower limb weakness). Magnetic resonance imaging evaluation was suggestive of acute infarct in the right frontal lobe and basal ganglia. Right middle cerebral artery thrombosis (►**Fig. 8**). The

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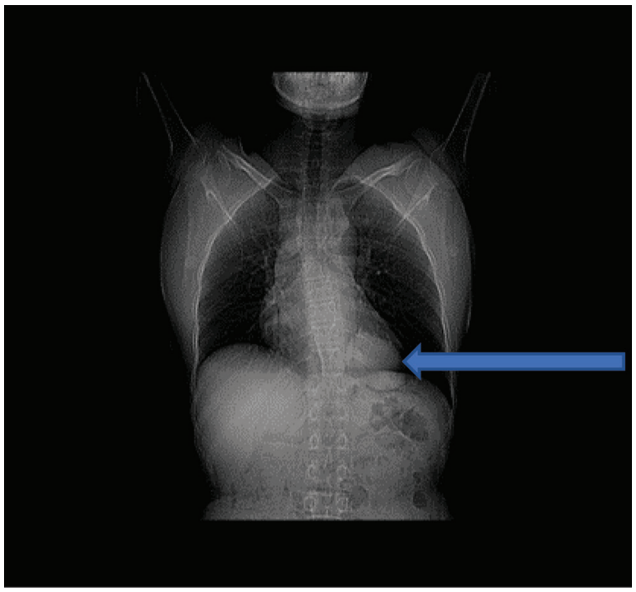

Fig. 1 Chest X-ray: well-defined opacity in the left lower zone near the cardiophrenic region.

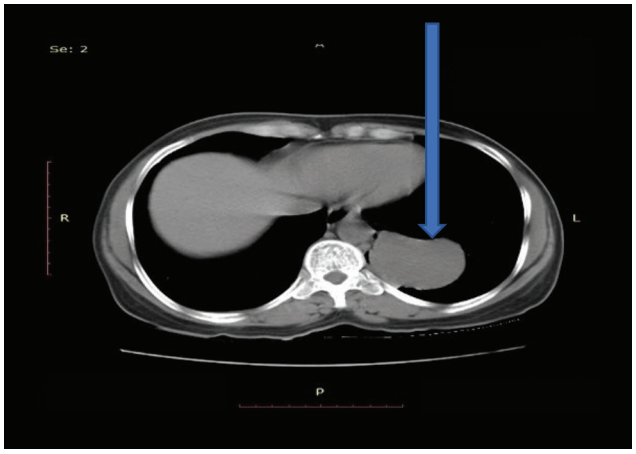

Fig. 2 Computed tomography thorax: hypodense cystic lesion in the posterior basal region of the left lobe.

patient was further evaluated for the cause of the cerebrovascular accident, and on evaluation with echocardiography, it was suggestive of left PVT extending to the left atrium.

Treatment

The patient was started on anticoagulation therapy along with antiplatelets and with regular physiotherapy. On follow-up after 1 month of treatment initiation, the patient has regained partial mobility of the left upper and lower limb.

Discussion

Patients undergoing partial or complete lung lobe resection are prone to develop PVT. Following lobectomy, the risk of PVT is 3.6%, which elevates to 13.6% in those undergoing left upper lobectomy[3,4,5]. Signs and symptoms include dyspnea, hemoptysis, chest pain, fever, and hypoxemia. In severe cases, the patient may develop a transient ischemic attack

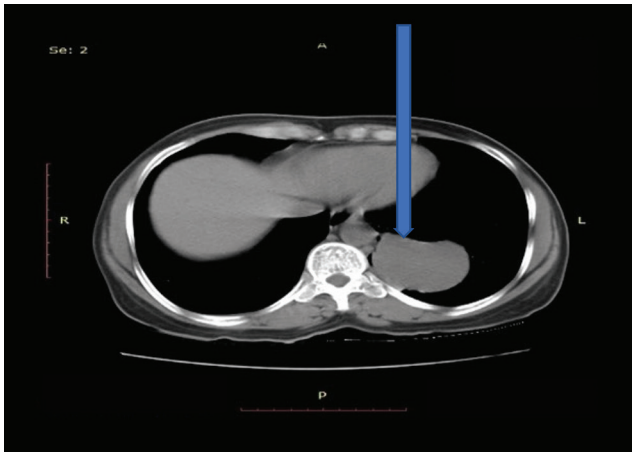

Fig. 3 Contrast enhanced computerized tomography thorax: well-defined rounded, hypodense, thin-walled nonenhancing uniloculated lesion—posterior basal segment of the left lower lobe. No surrounding consolidation or associated lesions.

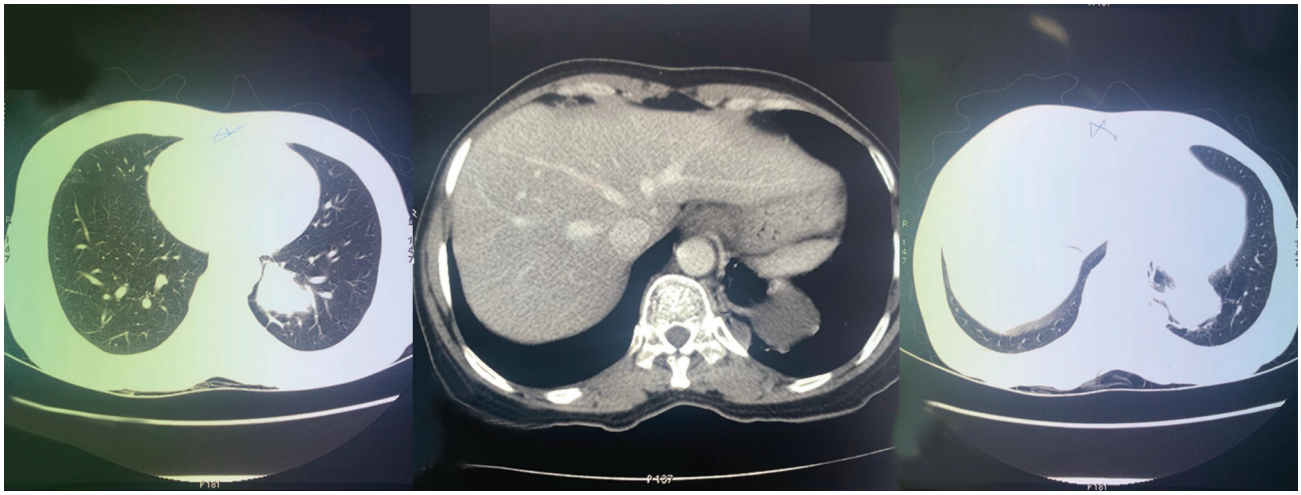

Fig. 4 Contrast enhanced computerized tomography thorax: vascular supply—two small arterial branches arising from descending thoracic aorta supplying the lesion and venous drainage into the pulmonary circulation.

Indication : Left lower lobe cystic leision
Route : Nasal
Vocal cords : Normal
Trachea : Normal
Main Carina: Normal
Right Bronchial Tree: Normal
Left Bronchial tree: Normal
No Endobronchial communication noted.
Biopsy : Not taken
Impreciation: Normal endobronchial airway
Cystic leision is intra lobar bronchopulmonary sequenstration.

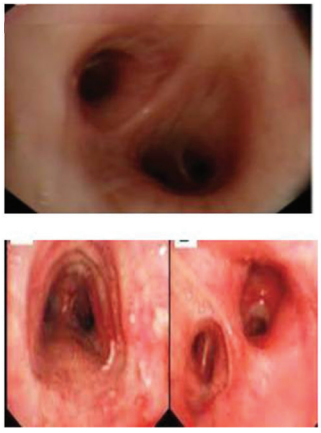

Fig. 5 Bronchoscopy: no endobronchial communication suggestive of intralobar sequestration.

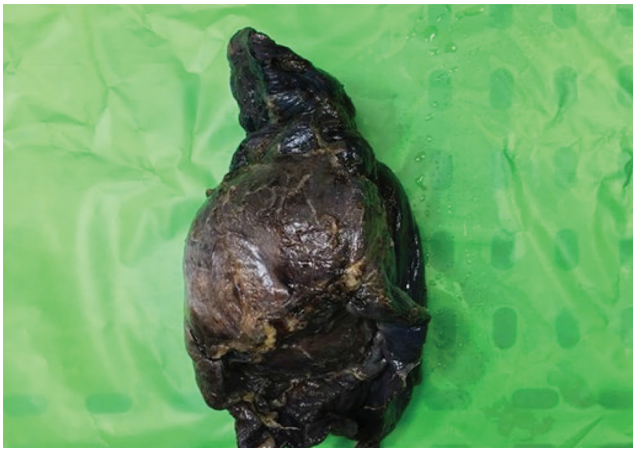

Fig. 6 Resected left lower lung lobe.

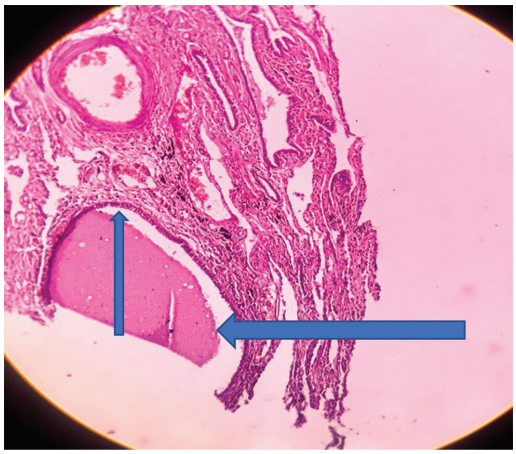

Fig. 7 Cyst lined by ciliated columnar epithelium having a uniform nucleus, cyst contains serous secretions.

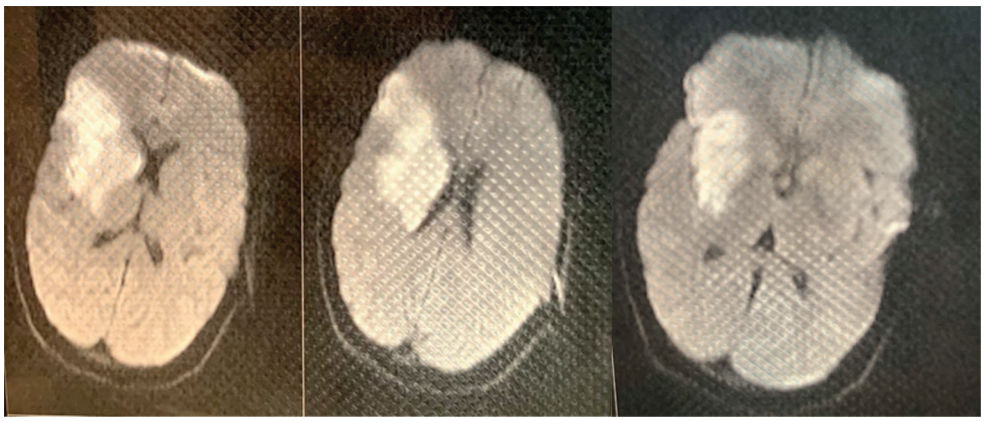

Fig. 8 Magnetic resonance imaging brain: acute infarct in the right frontal lobe and basal ganglia. Right middle cerebral artery thrombosis.

or a stroke. In the case reported here, the patient developed PVT after left lower lobectomy which has not yet been published in the literature to the best of our knowledge. In available literature, there have been only six reported cases of PVT after lobectomy.[6] All these cases involved resection of the left upper lobe with subsequent thrombus formation in the left upper PV stump. The true incidence of PVT after lobectomy is unknown and likely to be underdiagnosed, especially

in asymptomatic patients who do not undergo postoperative imaging with transesophageal echo (TEE) or computerized tomography (CT).

There is no sufficient literature evidence to suggest the routine use of postprocedure prophylactic anticoagulation or routine imaging and echocardiography in patients undergoing lung lower lobectomy.

CCAM is a congenital abnormality of the lung, which is uncommon in adults. Most CCAM involves unilateral lobes of the lung and may be complicated with pulmonary bacterial infections and abscesses. CCAM accompanied by Aspergillus infection may occur in bilateral lobes of the lung. The clinical and radiological findings vary, thus pathological examinations are required to make a definite diagnosis of cystic lesions of the lung. Type 2 CCAM represents up to 25% of all congenital lung lesions, and the vast majority are diagnosed within the first 2 years of life and occasionally present later in childhood but is a rare entity in adulthood.

Conclusion

In conclusion, the present case has showed that thrombus in pulmonary vein stump which is common after left upper lobectomy can still happen/occur after left lower lobectomy. Hence, it is recommended also to have routine echocardiographic evaluation (and CECT thorax if necessary) for patients undergoing left lower lobectomy in the same way as it is customarily done for patients with left upper lobectomy.

If cystic lesions are found in lower lobes of lungs, CCAM should be considered in the differential diagnosis even in adults even though they are rare.

A multicentered clinical study is advisable to elucidate the causes, frequency of PVT and also to evaluate and validate anticoagulant therapy for PVT following left lower lobectomy.

Conflict of Interest
None declared.

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