Idiopathic Bilateral Segmental Pulmonary Artery Aneurysm

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Abstract

Aneurysms of segmental pulmonary arteries are rare. They can be potentially fatal, but remain undetected in the majority of cases. A young person reported with massive hemoptysis, and a ruptured large segmental pulmonary artery aneurysm of the left lung was identified as the cause. Incidentally a smaller aneurysm on the segmental artery of the right lung was noticed. The case is of rare occurrence, and the urgency of intervention on the left lung with consideration of an aneurysm on the right lung makes this case challenging. The clinical decision making for the choice of procedure for this rare and emergent situation is presented.

Comment

Aneurysm in the pulmonary artery is a rare incidence. Commonly they are witnessed in the main or branched pulmonary artery segments. Their occurrence in a segmental artery is rare. The usual reason is a prior intervention in the pulmonary artery or association with a congenital cardiac abnormality, tumor, embolization, fungal infection, or cystic medial necrosis. Pseudoaneurysm is also reported after an unsuspecting pulmonary artery catheterization [4, 5]. A true idiopathic occurrence is extremely rare, and further, its bilateral presence and urgency of intervention in this patient made it unique as well as a challenging clinical decision to choose the appropriate management strategy.

Pulmonary artery aneurysms are known to be asymptomatic for a long period, and their discovery may be incidental. An indication to intervene is if the person has a history of hemoptysis or the size is more than 6 cm [6]. The smaller asymptomatic aneurysm is known to grow in size with time, as explained by the law of Laplace [7]. The SPAA on the left side had ruptured into the bronchus and

The branches of the left apical segment arose just before the origin of the aneurysm (Fig 1). There was another aneurysm of 2.5 cm arising from the lower medial basal segmental artery (Fig 1). There was evidence of air in the aneurysm of the left side indicating its rupture into the bronchus (Fig 1). Biochemical investigations were within normal limits with a hemoglobin level of 6 g/dL. Pulmonary function tests were within expected range for age and body surface area. Two units of packed red blood cells were transfused before the operation.

A left pneumonectomy was performed under general anesthesia. Postoperative recovery was normal, and ambulation was accomplished on the third postoperative day. After a week of near-normal activities with minimal effort, a catheter-based intervention was planned for the right SPAA. The procedure was performed under general anesthesia with endotracheal intubation. Coil occlusion of the right SPAA was achieved until its origin from the medial basal segmental pulmonary artery was reached (Fig 2). After the procedure, no flow in the artery and into the aneurysm was witnessed. Extubation was performed after 12 hours of elective ventilation.

The patient was ambulatory on the day of extubation. Postintervention recovery was unremarkable. There was no symptom of chest pain on the right side, dyspnea, or hemoptysis. The parenchyma of the right lung was normal on chest roentgenograph. The patient was discharged from the hospital on the tenth postoperative day.

Histopathologic examination confirmed the idiopathic nature of the aneurysm. The lung parenchyma and the unaffected pulmonary artery segments were of normal architecture. A section from the aneurysm shows fibrointimal hyperplasia and excessive fibrinoid deposition with fragmentation of the internal elastic lamina (Fig 3). These changes were commensurate with the presence of increased wall stress in the aneurysmal segment.

Ocurrence of aneurysm in the pulmonary artery system is rare. Most of these are congenital and associated with other intracardiac anomalies. Other reasons may be tumor, embolization after a procedure on the pulmonary artery, fungal infection, and cystic medial necrosis [1]. Usually they are reported in main and branched pulmonary arteries, but there are a few reports of its evidence in segmental arteries [2]. Although rare, this condition is potentially fatal [3]. We report a rare case of a segmental pulmonary artery aneurysm (SPAA) present in both lungs. Various clinical issues in management of this case are discussed.

A teenager presented to the emergency room with the complaint of massive hemoptysis. Approximately 300 mL of blood loss was estimated after a violent bout of coughing. After admission, hemodynamic stabilization was achieved. There was a history of the presence of scanty fresh blood in white and thick expectoration for the past 1 month. There was no past history of fever, dyspnea, chest pain, or trauma. There was no history of tuberculosis or hypertension in the past either. Chest roentgenograph revealed a large, spherical radiopaque shadow at the hilum of the left lung and a small radiopaque shadow in the parenchyma of the lower portion of the right lung. A contrast computed tomographic scan was performed, which revealed an 8-cm spherical aneurysm arising from the main lower segmental artery on the left side (Fig 1).

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thus needed urgent attention. This aneurysm was 8 cm in
diameter and arising from the segmental artery to the
whole of the lower lobe. The segmental arteries for
the apical lobe arose 8 to 10 mm before the origin of the
aneurysm (Fig 4). The other segmental arteries to the
segments of the lower lobe were arising from the lumen
of the aneurysm. These facts made the conservative
surgical or catheter-based intervention untenable. A
surgical strategy was planned to salvage the maximal
amount of lung tissue. The best option was to undertake a
left lower lobectomy. This was untenable because of the
fact that there was not enough clearance between the
origin of the apical lobar artery and the aneurysm so as to
safely divide and sew the pulmonary artery. Also, the size
of the aneurysm, especially when it is distended, will
make the hilar dissection for a lobectomy risky. Pneum-
onecctomy was a safe option that would produce the
least blood loss and fewest complications, so it was per-
formed on the left lung.

The right SPAA, although 2.5 cm and asymptomatic,
was destined to grow in size. Apart from this, in the
present situation it was limited to the medial basal
segmental artery, and any increase in size would have
affected other segmental arteries of the right lung.
Because the patient had only a right lung now, a con-
servative approach to treat this aneurysm to salvage the
maximal lung tissue was planned. The choice was to
salvage the complete right lung by attempting exclusion
of the SPAA by a covered stent [8]. This has been reported
frequently in main and branched pulmonary artery
segments but not further downstream. In our case, this possibility was ruled out as the proximal and the distal landing sites had a difference in diameter of 4 to 5 mm. The option of coil occlusion of the aneurysm from its origin in the right medial basal segmental artery was considered the best, and thus it was offered to the patient [9]. The patient is doing well postoperatively with no dyspnea or hemoptysis. Long-term follow-up of this case is required to observe the future course. Clinical follow-up every 6 months with computer tomographic angiogram, if required, is planned.

Histopathologic examination revealed a normal pulmonary artery architecture with no evidence of connective tissue disorder or lung disease. This qualifies as a true idiopathic aneurysm because the artery was dilated more than 30 mm with a saccular aneurysm on the right side, absence of intracardiac or extracardiac shunts, absence of chronic cardiac or pulmonary disease, absence of arterial wall disease, and normal pulmonary artery pressure [10]. Follow-up of this case is indeed required and we plan to perform it.

References