Late presentation of a pulmonary artery sling

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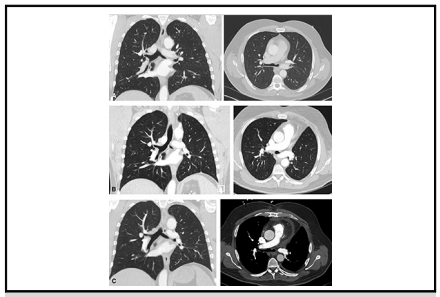

Radiologic progression of the bronchial compression at different surgical stages.

CENTRAL MESSAGE

Pulmonary artery sling is a rare congenital anomaly usually diagnosed in infancy. We present the case of an older patient whose anomaly became symptomatic after lung-reduction surgery.

Pulmonary artery sling (PAS) is a vascular ring anomaly in which the left pulmonary artery (LPA) arises from the right pulmonary artery (RPA) instead of the pulmonary trunk (PT). Symptoms including dyspnea, wheezing, and severe respiratory distress are nonspecific and mainly related to compression of the trachea by the aberrant LPA. Patients undergoing surgical repair of a PAS usually are children, ranging in age from 6 days to 27 months.[E1] We report the case of a 56-year-old patient whose PAS became symptomatic after a left upper lobectomy (LUL) for a solitary pulmonary nodule.

CASE PRESENTATION

A 56-year-old male patient underwent computed tomography (CT) of the chest for hemoptysis, with the incidental finding of a pulmonary carcinoid (pT1aN0), for which he underwent a LUL when indicated. On preoperative CT, the PAS was demonstrated, but surgical correction was not indicated, as the patient was asymptomatic. After his lobectomy, he developed worsening exertional dyspnea, which was investigated. A new CT of the chest demonstrated an increased compression of the trachea and the left main bronchus from the aberrant RPA (Figure 1). The decision was made to undergo surgical repair of the anomaly to relieve symptoms.

The patient was brought in the operating room and the usual anesthetic preparation was made, including a bronchoscopy. A median sternotomy was performed and the pericardium was opened. The aorta and right atrium were canulated, and cardiopulmonary bypass (CPB) was started. The LPA originated from the RPA, and its retrotracheal course was dissected under CPB and then both pulmonary arteries were clamped when well visualized. The LPA was closed with a polypropylene suture and then an anastomosis on the PT was performed with a 4-0 polypropylene suture. Weaning off CPB was uneventful, and the incision was closed by the standard method.

The postoperative course was uneventful, and patient left the hospital after 7 days. He developed a 9-mm segmental pulmonary embolism in the right upper lobe 9 days after surgery and severe acute respiratory syndrome coronavirus 2 infection 1 month after surgery. At 6 weeks' follow-up, the CT of the chest demonstrated resolution of the embolism in the right upper lobe. The patient is doing well, with exertional dyspnea that is still improving 6 months' postoperatively. The patient provided written consent for publication of their study data; institutional review board approval was not required.

DISCUSSION

We report a rare case of a PAS, which became symptomatic after a left upper lobectomy for a carcinoid tumor. PAS are usually diagnosed in infants, and the median age for surgical correction is 7 to 9 months.[E2] A total of 29 adult patients diagnosed with this anomaly are reported in the literature, to the best of our knowledge, from whom only 2 underwent surgery.[1-4] The first patient underwent surgery for asthma and severe dysphagia associated with tracheal and esophageal compression by the aberrant RPA, whereas the second one underwent a simultaneous lung resection and correction of the anomaly.[3,4]

Although the case reported by Mammana and colleagues[4] reports the simultaneous correction of the PAS

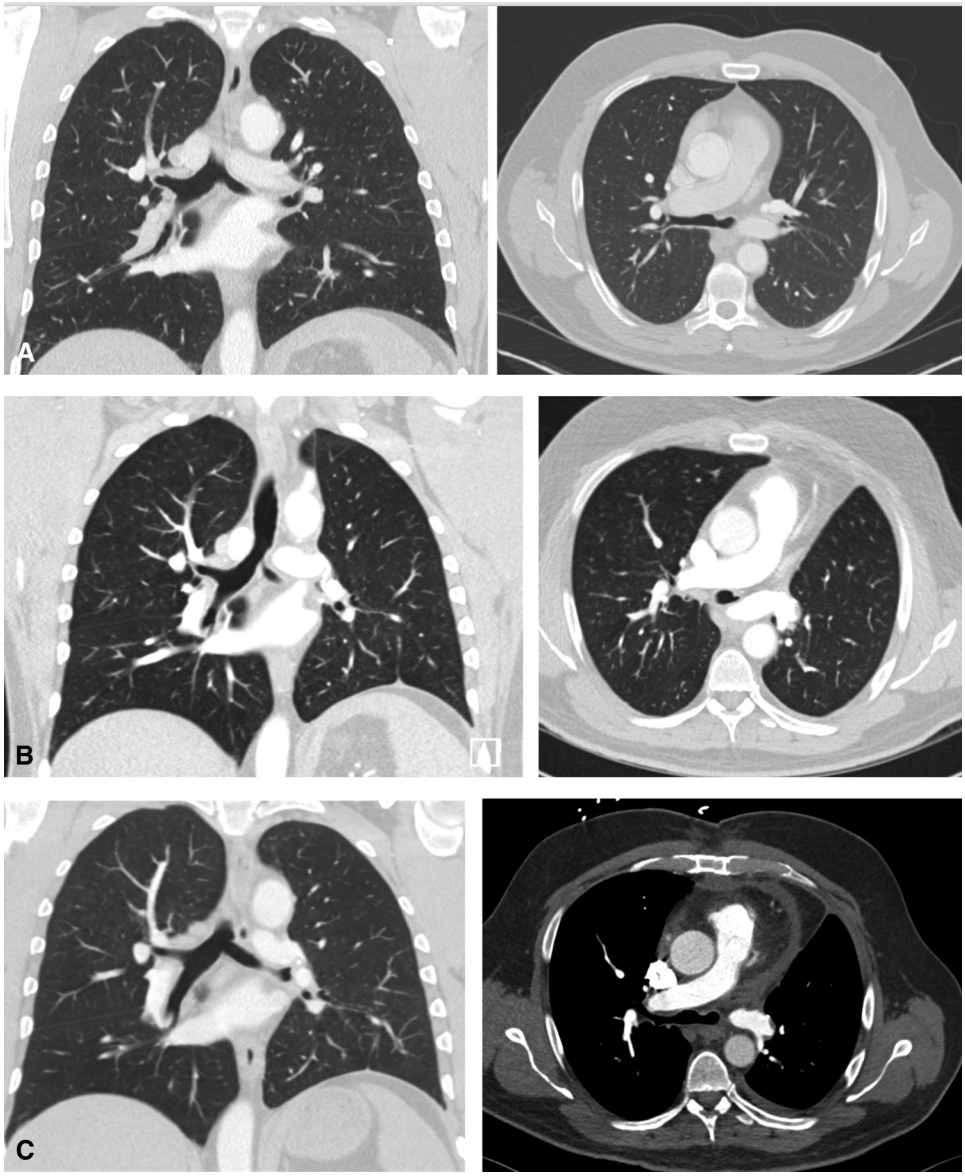

FIGURE 1. Radiologic progression of the bronchial compression. Computed tomography (CT) of the chest demonstrates a decreased size of the left main bronchus from 9 mm previously (A) to 5 mm after lung surgery (B). The CT after cardiac surgery (C) demonstrates an improved diameter of the left main bronchus at 7 mm.

in a patient with a lung nodule, their patient had vascular invasion by the adenocarcinoma, which justified the one-stage surgery. In our patient, the patient was asymptomatic from his PAS, and the lung nodule was smaller and did not invade the mediastinum and blood vessels; thus, PAS transposition was not judged indicated at time of the LUL. After the LUL, the mediastinum shifted towards the empty space in the left pleura and increased the tension created by the LPA on the right main bronchus. The native asymptomatic stenosis then became clinically significant with increased exertional dyspnea, thus suggesting that surgical correction should be undertaken. The surgical correction of PAS in children is well described, and the technique includes CPB with mild hypothermia and beating-heart.[E2,5] Then, the LPA can be dissected from its retrotracheal course and dissected near its origin and re-implanted on the PT.[E2,5]

Our case demonstrates that lung-resection surgery on the contralateral side of the aberrant pulmonary artery could increase tracheal compression. Thus, an asymptomatic PAS could become symptomatic after resection and surgery can be delayed, allowing recovery from one surgery before the other. If the lung resection is performed on the same side as the aberrant pulmonary artery, then lobectomy should not

increase the stenosis and create new symptoms, as the mediastinum would shift on the opposite side.

CONCLUSIONS

This is the first case of a staged surgery for the resection of a pulmonary carcinoid tumor and subsequent correction of the PAS. The favorable outcomes suggest that surgery could be delayed to allow the patient to recover from thoracic surgery before undergoing cardiac surgery.

Conflict of Interest Statement

The authors reported no conflicts of interest.

The *Journal* policy requires editors and reviewers to disclose conflicts of interest and to decline handling or reviewing manuscripts for which they may have a conflict of interest. The editors and reviewers of this article have no conflicts of interest.

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