Superior Sulcus Tumor Resection With Multiple Pulmonary Arteriovenous Fistulas

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The authors present a case of a 66-year-old male presenting with a superior sulcus tumor and severe hypoxemia due to bilateral multiple pulmonary arteriovenous fistulas (PAVF). The unilateral pulmonary arterial occlusion test was useful before surgery because it enabled evaluation of the feasibility and safety of intraoperative pulmonary artery clamp and one-lung ventilation during lung resection. Results facilitated safe resection of the superior sulcus tumor using the modified transmanubrial osteomuscular sparing approach, providing an excellent surgical field.


Bilateral multiple pulmonary arteriovenous fistulas (PAVF) are congenital abnormalities that produce hypoxemia through right-to-left shunts. Although coil embolization or surgical intervention can reduce the intrapulmonary shunt, improving oxygenation remains difficult in some cases. Significant hypoxemia can result from both decreased lung volume that occurs in lung resection and from intraoperative one-lung ventilation. This can result in adverse effects on circulatory dynamics and make it difficult to assess the feasibility of surgery. For high-risk patients, a unilateral pulmonary arterial occlusion (UPAO) test can guide the suitability of surgery by assessing predictive cardiopulmonary functions [1]. We report a case of superior sulcus tumor (SST) with bilateral multiple PAVF, wherein complete resection was safely performed using a transmanubrial osteomuscular sparing approach (TMA) after careful assessment with the UPAO test.

A 66-year-old male with a history of surgery for bilateral multiple PAVF presented with right chest pain. He had undergone surgery for PAVF at the age of 49 years (right, S3 segmentectomy; left, partial resection) and coil embolization at the age of 55 years. Clinically, there was cyanosis on the lips and nails, with marked clubbing of fingers. Despite complaining of mild dyspnea on exertion, blood gas analysis in room air revealed marked hypoxemia (PaO2 45.6 mm Hg). Chest computed tomography (CT) revealed a mass shadow in the apex of the right lung, measuring 39 × 23 mm in diameter that had invaded the first rib (Fig 1A), with continuing evidence of bilateral PAVF despite his earlier treatment (Fig 1B). A definitive diagnosis of non-small cell lung cancer was achieved using percutaneous CT-guided biopsy, and radiation therapy was initiated because of no mediastinal lymph nodes swelling on chest CT.

A ventilation-perfusion scintigraphy was requested, which revealed an intrapulmonary right-to-left shunt of 35.9%, with 61% right lung perfusion and 39% left lung perfusion. Coil embolization was performed 2 times for relatively large arteriovenous fistulas in the left lung, aiming to improve the hypoxemia. However, the intrapulmonary right-to-left shunt ratio and blood gas analyses remained unchanged.

Respiratory function testing revealed decreased forced expiratory volume in 1 second (FEV1) of 1.4 (61.6% of predicted), decreased FEV1/forced vital capacity ratio of 36.18%, and decreased diffusing capacity of lung for carbon monoxide (DLCO) of 8.90 (58.3% of predicted). Despite this severe obstructive pattern, predictive postoperative (ppo) results exceeding 40% for both %ppoFEV1 and %ppoDLCO revealed that surgery may be feasible. Therefore, a UPAO test was performed to predict cardiopulmonary function and to provide further reassurance on the safety of surgery. Briefly, a thermodilution catheter with a balloon was inserted through the femoral vein to measure the mean pulmonary artery pressure (mPAP), the cardiac index (CI), and the total pulmonary vascular resistance (TPVRI) before occlusion and during 15 minutes of occlusion. The CI was measured by the thermodilution method, and TPVR was calculated from the mPAP and CI. In the condition of right main PA occlusion, mPAP, CI, and TPVRI were 26 mm Hg, 4.04 L · min⁻¹ · m⁻², and 514.3 dyne · sec · cm⁻⁵ · m²⁻¹ respectively. On the basis of these data, we decided to perform radical pulmonary resection.

The surgery was performed with the patient in the supine position, using two-lung ventilation for as long as is safely possible. We used TMA associated with a median sternotomy and this approach provided an excellent surgical field. The subclavian vein was easily detached from the tumor by simple dissection, and the first rib and intercostal muscle were excised with sufficient surgical margin from the infiltrating portion of the tumor. The entire right lung adhered to the chest wall and the hilum was obliterated by dense adhesions because of the prior S3 segmentectomy. Despite these challenges, right upper lobectomy and upper mediastinal lymph node dissection were successfully performed.

Postoperative pathologic examination confirmed the diagnosis of pulmonary adenocarcinoma and that the surgical margins were free from cancer. The tumor was staged according to the Union for International Cancer Control as ypT3N0M0; stage IIB. Even though the patient’s recovery was uneventful, he was discharged from hospital with home oxygen therapy on the 13th post-operative day because of marked hypoxemia (PaO2 38.3 mm Hg) on postoperative blood gas analysis in room air. The PaO2 was 53.6 mm Hg and partial pressure of carbon dioxide, arterial (PaCO2) was 43.9 mm Hg under

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administration of 3 L/minute oxygen; initial symptom of moderate dyspnea on exertion had gradually reduced on subsequent clinical course. Over a 22-month follow-up period, there has been no evidence of recurrence.

**Comment**

Our patient presented with severe hypoxemia due to multiple right-to-left shunts. Despite coil embolization of relatively large PAVF in the left lung, there was no improvement in oxygenation.

Although preoperative lung function testing revealed a markedly obstructive pattern, both diffusion and ventilatory capacity presented average risks for lung resection with both %ppoDLCO and %ppoFEV1 greater than 40% [2]. However, pulmonary perfusion scintigraphy revealed higher perfusion in the right lung. Therefore, complications were anticipated in the perioperative period because of a potential inhibitory effect on left ventricular function or an increase in pulmonary vascular resistance. This could have resulted from the smaller pulmonary vascular bed that accompanied the patient’s severe emphysema, the lung resection, or the intraoperative hypoxemia resulting from the pulmonary artery clamp or one-lung ventilation. Therefore, we decided to perform the UPAO test to evaluate simulations using the pulmonary artery clamp or one-lung ventilation.

The UPAO test assessed the predictive postoperative cardiopulmonary function and the lung resectability in our patient with compromised pulmonary function. Using the UPAO test, tolerance limits for lung resection have been reported as TPVRI of less than 700 dyne · sec · cm⁻⁵ · m² and mPAP of less than 30 mm Hg [1]. In the present case, mPAP, CI, and TPVRI under right main pulmonary artery occlusion were within tolerance limits for a radical pneumonectomy. Therefore, we decided to perform the UPAO test to evaluate simulations using the pulmonary artery clamp or one-lung ventilation.

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A TMA is a recognized approach for the surgical management of SST and has been improved by Grunenwald and Spaggiari [3], providing an excellent surgical field and scope for extended resections while avoiding postoperative deformities of the sternoclavicular joint. In our case, with SST situated in the anterior inlet, we selected TMA because of the need for an uncompromised view of subclavian vessels during the combined resection of the first rib and the need for careful detachment of the tumor from the right subclavian vein. Furthermore, we anticipated strong adhesions around the pulmonary hilum and chest wall because of the previous segmentectomy of the right lung for PAVF. Consequently, we used a median sternotomy approach to ensure the right main pulmonary artery and associated vessels could be safely manipulated. We considered that combined with TMA, this approach provided good exposure of the surgical field under safer conditions.

In conclusion, we report the successful resection of SST in a case complicated with bilateral multiple PAVF. We believe that the UPAO test contributed to successful surgical outcome by predicting the possible postoperative cardiopulmonary function and providing surgical simulations.

**References**