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Partial Anomalous Pulmonary Venous Return in Lung Cancer Surgery: Diagnostic Challenges and Surgical Considerations in Two Cases

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Partial anomalous pulmonary venous return (PAPVR) is a rare congenital anomaly in which one or more pulmonary veins drain into the systemic venous system instead of the left atrium, in the absence of associated cardiac defects such as atrial septal defect. When only a single anomalous pulmonary vein is involved, the resulting shunt volume is typically small, allowing asymptomatic progression into advanced age. Consequently, such cases are often incidentally diagnosed or remain undetected. We report herein two cases of PAPVR that required pulmonary resection for primary or metastatic lung cancer. In Patient 1, PAPVR was successfully identified preoperatively using three-dimensional computed tomography in the lobe with cancer, enabling appropriate surgical planning for lobectomy. In Patient 2, segmentectomy was performed to preserve lung function: PAPVR was not recognized preoperatively on conventional two-dimensional computed tomography but only identified postoperatively, in a different lobe to the one with cancer. A deliberate awareness of the possible presence of PAPVR and accurate identification of PAPVR before surgery are essential in patients undergoing pulmonary resection. Segmentectomy may be a viable surgical option in select cases complicated by PAPVR because it potentially minimizes the reduction in pulmonary blood flow while preserving lung function.

Key words: computed tomography, lobectomy, lung cancer, partial anomalous pulmonary venous return, segmentectomy

INTRODUCTION

Partial anomalous pulmonary venous return (PAPVR) is a rare congenital anomaly in which one or more pulmonary veins drain into the systemic venous circulation instead of the left atrium, while at least one pulmonary vein maintains normal drainage. Its reported prevalence ranges from 0.4% to 0.7% [1, 2]. In isolated cases without associated cardiac anomalies, a single anomalous vein typically results in minimal left-to-right shunting; such cases are often asymptomatic and they are extremely rare [3]. With the increasing use of high-resolution imaging modalities such as contrast-enhanced computed tomography (CT) and three-dimensional reconstruction, incidental detection of PAPVR has become more common, particularly during preoperative evaluation for lung cancer. Although asymptomatic, PAPVR can significantly influence surgical planning, especially in cases in which pulmonary resection is required. In these cases, undetected anomalous venous return may lead to unexpected abrupt hemodynamic changes during surgery. We report two cases of PAPVR that was incidentally identified during preoperative imaging for lung cancer. In one case, the anomaly was overlooked preoperatively and recognized only after surgery, confirming the importance of meticulous preoperative assessment and the clinical implications of venous anomalies in thoracic surgical practice.

CASE PRESENTATIONS

Patient 1

A 73-year-old woman without symptoms was referred to our department after an abnormal shadow was detected in the right lung field on a chest radiograph during a routine medical checkup. She was a nonsmoker with a history of dyslipidemia; a physical examination revealed no remarkable findings. CT

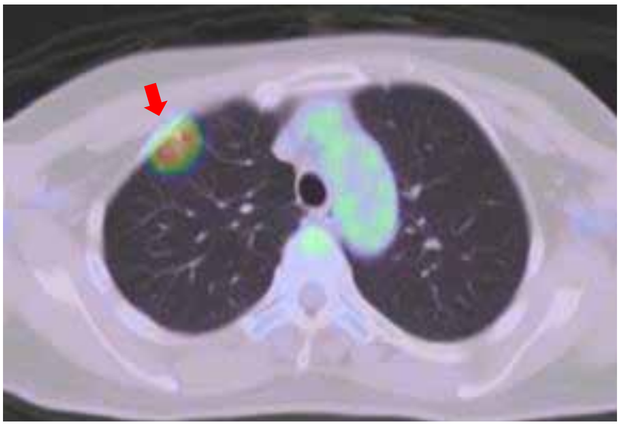

Fig. 1 Positron emission tomography image showing distinct fluorodeoxyglucose uptake.

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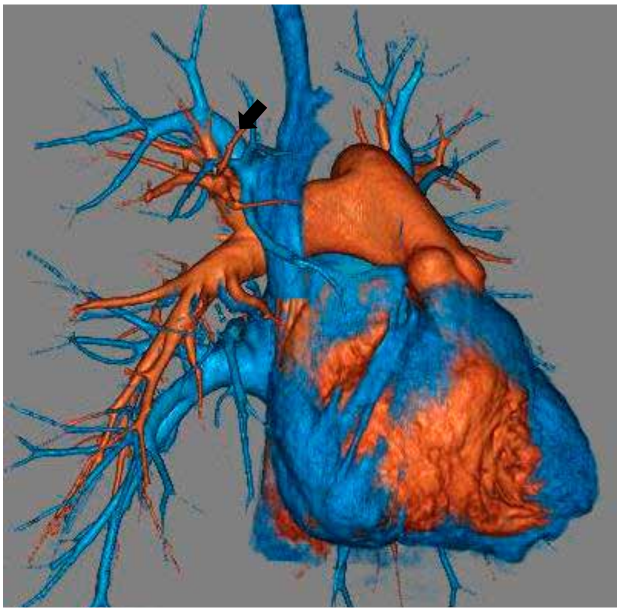

Fig. 2 Three-dimensional computed tomography image revealing an anomalous right upper pulmonary vein draining into the superior vena cava (black arrow).

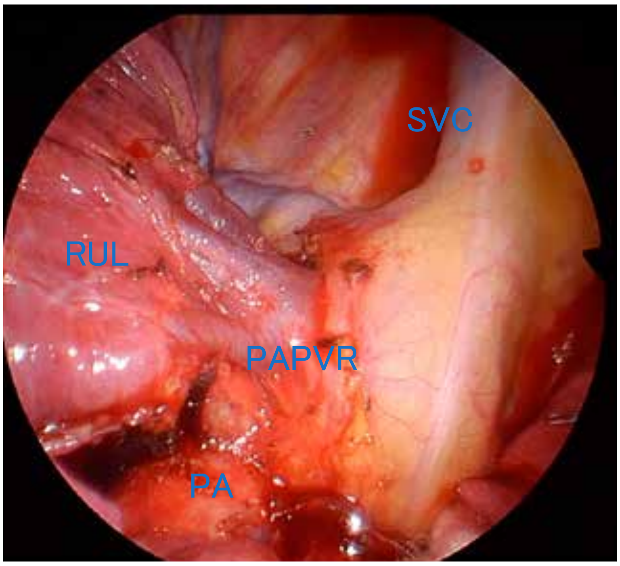

Fig. 3 Anomalous venous drainage into the superior vena cava is observed under the thoracoscopic view. PA, pulmonary artery; PAPVR, partial anomalous pulmonary venous connection; RUL, right upper lobe; SVC, superior vena cava.

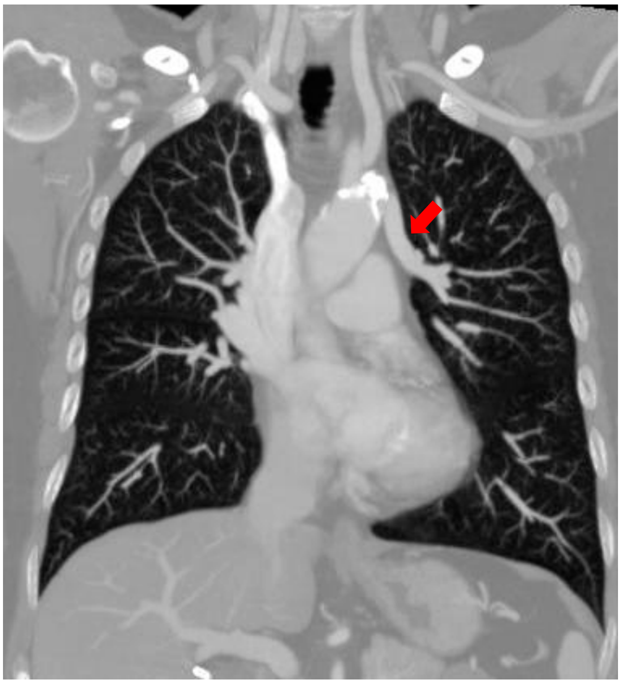

Fig. 4 Two-dimensional computed tomography image revealing an abnormal venous return in which the pulmonary vein of the left upper lobe drains into the left brachiocephalic vein (red arrow).

revealed a 30-mm part-solid ground-glass nodule in the right upper lobe. The ground-glass component of the lesion was predominant, suggesting relatively low tumor cellularity; however, positron emission tomography showed distinct fluorodeoxyglucose uptake (maximum standard uptake value: 6.2) with no evidence of lymph node involvement or distant metastasis (Fig. 1). The clinical diagnosis was early-stage invasive lung cancer, and surgery was planned. Three-dimensional CT (3D-CT) revealed an anomalous right upper pulmonary vein draining into the superior vena cava just below the azygos vein (Fig. 2). The pulmonary vein of the middle lobe returned independently to the left atrium. Preoperative echocardiography confirmed normal cardiac function with no signs of right atrial overload, right ventricular enlargement, or pulmonary hypertension. Anomalous venous drainage into the superior vena cava was confirmed under thoracoscopic guidance (Fig. 3). The anomalous vein, upper-lobe pulmonary artery, and bronchus were sequentially dissected and a right upper lobectomy with lymph node dissection was performed. Contrary to expectations, histological analysis confirmed the presence of minimally invasive adenocarcinoma. The patient's recovery was uneventful and she was discharged on postoperative day 4. A follow-up examination was conducted 2 years and 9 months after surgery: No signs of recurrence were noted, and echocardiography confirmed that cardiac function remained stable with no evidence of right heart strain. The patient remained asymptomatic and was followed through routine oncological surveillance.

Patient 2

A 72-year-old woman was referred to our department after a pulmonary nodule was detected on CT images. The CT examination was performed during follow-up evaluation after surgical treatment for colon cancer. The patient was a nonsmoker with a history of hypertension. CT revealed a 19-mm nodule in segment 6 (S6) of the left lower lobe; which had increased in size compared to imaging findings from 6 months earlier. Based on clinical evaluation, the lesion was suspected to be a metastatic tumor arising from the colon cancer. Thoracoscopic left S6 segmentectomy, including part of S10, was performed. Histological analysis confirmed metastatic adenocarcinoma originating from the colon. The patient's postoperative course was uneventful, and she was discharged on day 5. Reevaluation of the initial preoperative imaging, using two-dimensional CT (2D-CT), revealed an abnormal venous return in which the pulmonary vein of the left upper lobe drained into the left brachiocephalic vein (Fig. 4). Preoperative echocardiography and electrocardiography revealed normal cardiac function. A reassessment of cardiac function was performed, and both echocardiography and electrocardiography showed normal cardiac function with no evidence of associated cardiac anomalies. Although the abnormal return of the upper pulmonary vein persisted, no cardiac dysfunction was observed at 2 years and 3 months postoperatively. This case has been previously reported in two separate publications: one focusing on the anatomical characterization of the pulmonary venous anomaly [4], and the other on the technical aspects of segmental resection using multidetector CT angiography [5]. In the present study, we revisit this case in the context of PAPVR-associated lung cancer, emphasizing long-term hemodynamic stability and surgical implications. Unfortunately, the patient passed away 3 years later due to an unrelated illness.

DISCUSSION

We analyzed 18 cases of PAPVR associated with lung cancer, including one case of metastatic tumor, that have been reported since 2013, including two our current cases (Tables 1 and 2) [5–20]. In our cases, PAPVR was incidentally detected during perioperative evaluation for lung cancer; both cases were of the isolated type with no intracardiac anomalies and a single anomalous vein. An anomalous vein is typically asymptomatic, but its presence in a non-tumor lobe

Table 1 Tumor and PAPVR located same lobe

Case	Age	Sex	Procedure	PAPVR Site	Preoperative Diagnosis	Qp/Qs	Repair	Complication
1 (Present case)	73	F	RULx	RUL → SVC	Yes	N/A	No	None
2 (Kurihira[11])	64	M	RULx → LULx	RUL → SVC	Yes	N/A	No	None
3 (Isaka[8])	70	M	RULx	RUL → SVC	No	N/A	No	None
4 (Kawasaki[13])	45	M	RULx	RUL → SVC	Yes	1.08	No	None
5 (Tamburini[15])	54	F	RULx	RUL → SVC	No	N/A	No	None
6 (Inafuku[16])	59	M	RULx	RUL → SVC	Yes	N/A	No	None
7 (Liu[17])	48	M	RPx	RLL → IVC	Yes	N/A	No	None
8 (Asakura[18])	64	M	LULx	LUL → BCV	Yes	2	No	None
9 (Akabane[10])	73	M	LS1 + 2 + 3seg and RULx symoutansous resection	RUL → SVC	No	N/A	No	None
10 (Singhal[12])	70	M	R S3 seg	RUL → SVC	Yes	1.4 → 1.3	No	None
11 (Muranishi[20])	63	F	LS1 + 2 + 3seg	LUL → BCV	Yes	1.12	No	None

Table 2 Tumor and PAPVR located different lobe

Case	Age	Sex	Procedure	PAPVR Site	Preoperative Diagnosis	Qp/Qs	Repair	Complication
12 (Present case)	72	F	LS6 seg	LUL → BCV	No	N/A	No	None
13 (Yosimine[7])	78	M	RMLx	RUL → SVC	Yes	1.3 → 1.51	No	None
14 (Kurihira[13])	71	M	LLLx	LUL → BCV	Yes	1.14 → 1.3	No	None
15 (Matsui[6])	74	F	RLLx	RUL → SVC	Yes	0.98	No	None
16 (Fukumoto[9])	66	F	LULx + α	RUL → SVC	Yes	2.24	Yes (pre-op)	paf
17 (Kawagoe[14])	74	F	LLLx	LUL → BCV	No	N/A	Yes (intra-op)	None
18 (Nakata[19])	66	M	LLLx	LUL → SVC	Yes	N/A	Yes (intra-op)	None

RUL: right upper lobe, RULx: right upper lobectomy, RMLx: right middle and lower lobectomy, RLL: right lower lobe, RLLx: right lower lobectomy, RPx: right pneumonectomy, LUL: left upper lobe, LULx: left upper lobectomy, LLLx: left lower lobectomy, SVC: superior vena cava, IVC: inferior vena cava, Seg: segmentectomy, BCV: brachiocephalic vein, paf: paroxysmal atrial fibrillation, Qp/Qs: pulmonary-to-systemic flow ratio, N/A: not applicable poses a unique surgical challenge. If unrecognized, compensatory venous drainage may inadvertently be disrupted through resection, resulting in volume overload of the right heart. In Patient 1 (Case 1), an anomalous pulmonary vein that was confined to the tumor-bearing right upper lobe was identified preoperatively. A lobectomy using video-assisted thoracoscopic surgery was performed and no postoperative hemodynamic complications occurred. Patient 2 (Case 12) had a pulmonary metastasis from colon cancer in the left lower lobe and PAPVR in the left upper lobe was not recognized preoperatively due to limitations in imaging technology at the time. A segmentectomy was performed using video-assisted thoracoscopic surgery and right heart overload was successfully avoided. These cases highlight the importance of evaluating both the anatomical distribution and physiological impact of PAPVR when planning lung cancer surgery.

Historically, PAPVR that is associated with lung cancer more frequently occurs in the left upper lobe [14, 18], likely due to the relative ease of detecting a left-sided anomalous vertical vein on conventional axial CT imaging. However, our recent review of published cases (Tables 1 and 2) indicates a prevalence of right (61%) compared to left (33%) upper lobe PAPVR. The predominance of PAPVR in the right upper lobe draining into the superior vena cava closely parallels that of the most common subtype observed in pediatric cases. This anatomical consistency may reflect a shared embryological origin and suggests that such anomalies persist into adulthood. Therefore, right-sided PAPVR in patients with lung cancer may be more prevalent than previously considered. We hypothesize that this trend reflects the improved diagnostic sensitivity afforded by 3D-CT, which enables more accurate visualization of pulmonary venous anatomy and reduces the likelihood of right-sided anomalies being missed. In our review of 18 cases, PAPVR was diagnosed in 9 cases using 3D-CT, and remaining 9 cases using 2D-CT. In addition to carefully examining conventional 2D-CT, utilizing 3D-CT makes it possible to prevent overlooking vascular malformations.

From an educational standpoint, of note is that a Qp/Qs (pulmonary to systemic flow) ratio of 1.5 or greater is generally considered the threshold for surgical intervention in PAPVR because this value reflects a clinically significant left-to-right shunt. Although this threshold is often not reached in isolated single-vein PAPVR, the presence of PAPVR can still influence surgical strategy, particularly in oncological resections. Therefore, routine use of three-dimensional CT and, when indicated, Qp/Qs assessment should be integrated into preoperative planning to ensure both oncological and physiological safety. In the initial 11 cases, including that of Patient 1 (Case 1) of the present study, PAPVR was located within in the tumor-bearing lobe (Table 1). In these cases, the patients underwent either pneumonectomy, lobectomy, or segmentectomy with the aim of addressing both oncological and hemodynamic concerns. Surgical resection in this group appeared to confer oncological advantages and contributed to hemodynamic stability. For instance, a measurable reduction in pulmonary hypertension following lobectomy was demonstrated in Case 8, whereas a decreased Qp/Qs ratio was demonstrated in Case 10 following segmentectomy. Patient 1 (Case 1) remained clinically stable without signs of right ventricle overload at 2 years and 3 months after surgery. In contrast, seven patients, including Patient 2 (Case 12) of the current study, presented with PAPVR involving lobes that did not form part of the resected area (Table 2). Preoperative identification of these anomalous veins proved critical for ensuring postoperative hemodynamic stability. Of the seven patients with residual PAPVR, six underwent lobectomy and three required surgical repairs of the anomalous vein. One patient (Case 12) underwent segmentectomy. Among the three patients who underwent repair for PAPVR, one developed paroxysmal atrial fibrillation postoperatively, but the remaining six experienced no complications. These results suggest that surgical repair of an anomalous vessel is not always necessary in cases with low shunt volume and stable hemodynamics.

Notably, our patient (Case 12) underwent thoracoscopic segmentectomy of the left S6 region, with pulmonary veins V8 to V10 being preserved. Postoperative evaluation using echocardiography and electrocardiography at an examination 2 years and 9 months after surgery revealed no right heart strain. When PAPVR occurs in a different pulmonary lobe and the PAPVR remains, segmentectomy may preserve a greater proportion of physiological pulmonary venous return, potentially reducing the risk of postoperative hemodynamic instability. Although the number of segmentectomy cases is limited and individual anatomical variations must be considered, segmentectomy may offer advantages over lobectomy from an anatomical standpoint. These findings suggest that segmentectomy, when anatomically feasible, may represent a viable surgical option in selected cases of PAPVR-associated lung cancer.

CONCLUSIONS

Advances in imaging have enhanced diagnostic accuracy and surgical planning, leading to improved short-term outcomes. To prevent postoperative hemodynamic instability, particularly in cases involving anomalous pulmonary veins outside the resection area, a thorough preoperative assessment is crucial. Although PAPVR repair may be beneficial, safe outcomes are possible without repair when the shunt volume is low and venous return is preserved. When feasible, segmentectomy offers hemodynamic advantages because it enables normal venous return to be maintained; it may thus be a valuable surgical option.

CONFLICT OF INTEREST

There were no conflicts of interest.

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