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CASE REPORT

Pyogenic granuloma originating in the pulmonary artery

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Abstract
We herein report an unreported case of pyogenic granuloma that originated in the pulmonary artery. A 38-year-old man was urgently hospitalized with dyspnea and back pain. He had been on hemodialysis for 2 years due to chronic renal failure. We performed contrast-enhanced computed tomography and detected a mass occluding the left main pulmonary artery. The maximum standardized uptake value (SUV_{max}) of ^{18}F-fluorodeoxy glucose (FDG) in the mass was 4.1. We made a tentative diagnosis of pulmonary artery tumor, and planned an operation. We performed median sternotomy and left anterolateral thoracotomy. As the tumor had not reached the bottom of the left pulmonary artery, we first performed left upper lobectomy. We then performed resection of the pulmonary artery tumor under cardiopulmonary bypass and reconstructed the pulmonary artery with self-pericardium. The pathological diagnosis was pyogenic granuloma. To our knowledge, pyogenic granuloma originating in the pulmonary artery has never been reported before.

Keywords Pyogenic granuloma · Pulmonary artery tumor · Pericardial patch

Introduction

Pulmonary artery tumors are rare and almost always malignant. Given the similarities in the clinical and radiographic findings between pulmonary thromboembolism and pulmonary artery tumors in clinical and radiographic findings, it is difficult to make a conclusive diagnosis. Pyogenic granuloma is a well-known benign tumor. We herein report a case of a benign pulmonary artery tumor that was successfully resected under cardiopulmonary bypass with left upper lobectomy. To our knowledge, this is the first report of pyogenic granuloma originating in the pulmonary artery.

Case

A 38-year-old man was transferred to our hospital with dyspnea and left back pain. He had a medical history of dialysis treatment due to chronic glomerular nephritis. On admission, his pulse rate was 75/min, and his blood pressure was 138/75 mmHg. Physical examination, cardiac and lung auscultation were normal. There was no jugular venous distention, lymph node swelling or leg edema. The findings of a blood examination and serum tumor maker test were unremarkable. The respiratory function was within the normal limits. Deep leg vein echocardiography indicated no thrombus.

Chest contrast-enhanced computed tomography (CT) showed that the left main pulmonary artery was completely occluded by an enhanced homogeneous mass. Lymph node swelling was not detected (Fig. 1a). Lung perfusion scintigraphy indicated a lack of blood perfusion of the left lung. The maximum standardized uptake value (SUV_{max}) for ^{18}F-fluorodeoxy glucose (FDG) in this mass was 4.1. We made a tentative diagnosis of a benign pulmonary artery tumor, and planned an operation (Fig. 1b).

With the patient in the right half-lateral position, we performed median sternotomy and left anterolateral thoracotomy from the left fourth intercostal. As the tumor had not reached the left pulmonary artery A6, we concluded that the left lower lobe could be preserved. After upper lobectomy, the left pulmonary artery was incised under cardiopulmonary bypass. The tumor was 40 × 30 mm, elastic and soft with a smooth surface, and had a stem near Botalo's ligament (Fig. 2a, b). We resected the tumor along with part of the pulmonary arterial wall with a sufficient

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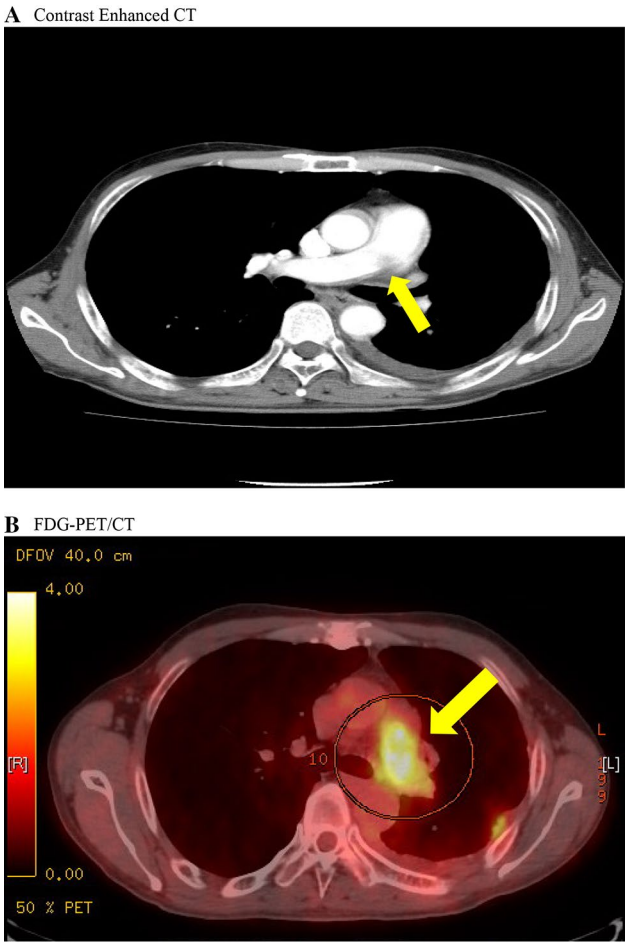

Fig. 1 Contrast-enhanced computed tomography shows that **a** a solid mass is completely occluding the left pulmonary artery and partially protruding into the main pulmonary artery (arrow). **b** An axial FDG-PET/CT image shows an increased FDG uptake in the part of the left pulmonary artery (SUV_{max} 4.1) (arrow)

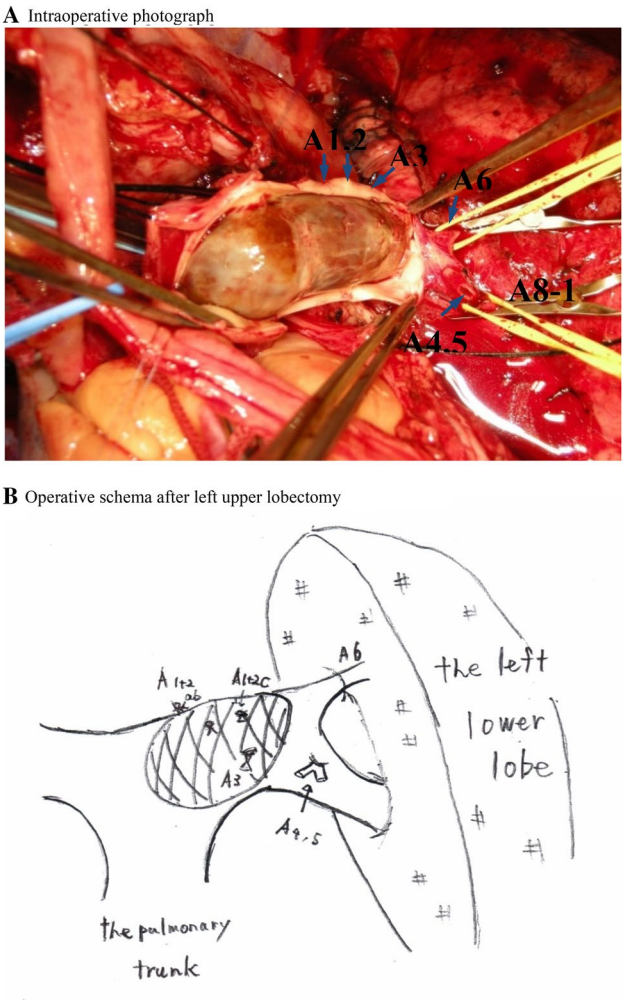

Fig. 2 **a** Intra-operative photograph. We incised the left pulmonary artery under cardiopulmonary bypass and detected a solid mass with a smooth surface extending near the left pulmonary artery A6. **b** A schematic illustration of the operative course after left upper lobectomy; the tumor did not reach the A6 with palpation

margin and reconstructed with a self-pericardial patch. The aortic cross-clamp time was 104 min, and the pump time was 141 min.

The post-operative course was uneventful. His symptom was improved. Post-operative contrast-enhanced CT of the left inferior pulmonary artery showed good visualization.

The gross surgical specimen was a single polypoid mass connected to the wall of the pulmonary artery. There were no necrotic or hemorrhagic lesions in the mass. A pathological examination revealed that the tumor consisted of round cells forming lobular proliferation of the capillaries. Endothelial cells were stained positive immunohistochemically for CD31/CD34 (Fig. 3a, b). Immunohistochemically staining also revealed Factor VIII and α-SMA positivity. Given these findings, a pathological diagnosis of pyogenic granuloma was made. The patient remains well with no evidence of recurrence five years after surgery.

Discussion

Primary pulmonary artery tumor is extremely rare, and while patients are typically asymptomatic for a time, the tumor begins to fill the pulmonary artery, eventually causing dyspnea. Contrast-enhanced CT, magnetic resonance imaging (MRI) and positron emission tomography (PET)/CT are useful for the differential diagnosis. In most previous reports of pulmonary artery tumor, the initial treatment method has been that for pulmonary thromboembolism (PTE), as it is frequently difficult to distinguish between pulmonary artery tumor and PTE [1].

In the present case, enhanced CT indicated an enhanced pulmonary mass. This suggested that the mass was not a thrombotic obstruction, as the thrombus would not be

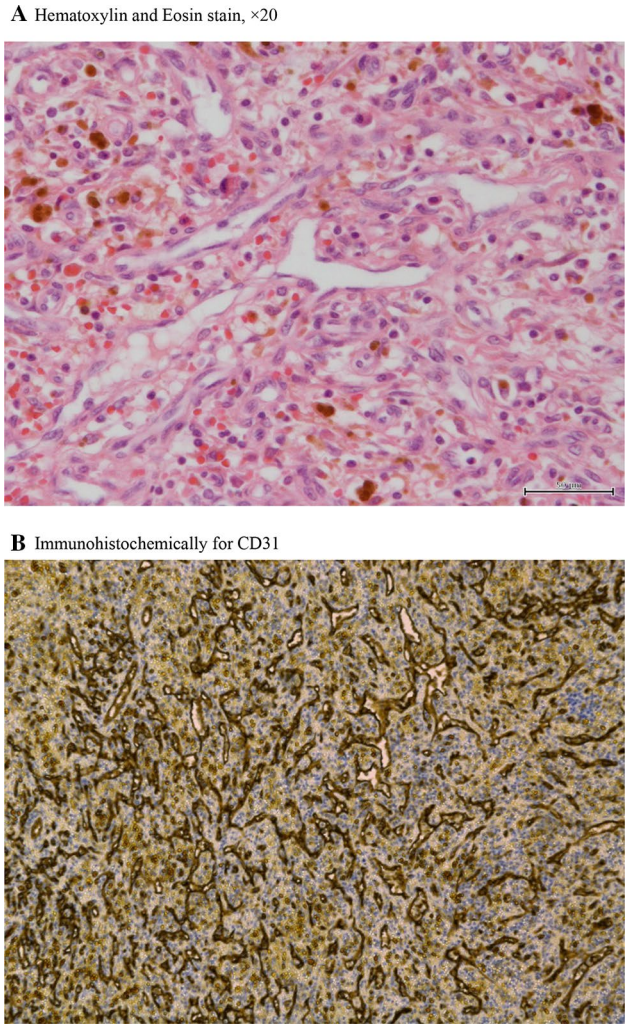

Fig. 3 The histopathological findings show. **a** Hematoxylin and eosin stain, ×20. The tumor consisted of round cells forming lobular proliferation of capillaries with inflammatory stroma. **b** Immunohistochemical findings for CD31, ×10. The immunohistochemical findings for CD31 were positive. The endothelial cells were increased in number and aggregated

enhanced. However, because we could not rule out PTE, we started anticoagulant therapy and performed other detailed examinations for the diagnosis. Malignant pulmonary artery tumors, such as sarcoma, are usually heterogeneously enhanced on CT, owing to the presence of necrosis, hemorrhaging and occasional ossification [1]. In contrast, benign tumors, such as myxoma, are homogeneously enhanced, so the enhancement can be useful for determining if the mass is benign or not [2]. Ito et al. suggested that FDG-PET might be useful for distinguishing PTE from pulmonary artery malignant tumors, such as pulmonary artery sarcoma (PAS). They reported the SUV_{max} for FDG in PAS was 7.25 ± 2.21, while that for thrombus was 2.36 ± 0.41; in the present case, the value was 4.1 [3].

Almost all reported cases of pulmonary artery tumor were diagnosed by post-operative pathological findings or autopsy findings because of the difficulty in achieving a pre-operative diagnosis. A CT-guided needle lung biopsy and transvenous catheter biopsy have been reported to be useful for confirming a diagnosis before operation [4]. However, a CT-guided needle lung biopsy is risky for proximal lesions and a transvenous catheter biopsy also carries a risk of distal embolism or tumor dissemination; we therefore did not perform these procedures before the operation. Whether the tumor is benign or not is the most important thing to determine during the operative procedure, if our hospital had possessed the appropriate technology, then we should have performed an intra-operative rapid pathological diagnosis.

Pyogenic granuloma (PG) is a well-known benign tumor that commonly occurs as a reactive lesion of the oral cavity or upper extremities. The main etiology for PG is said to be chronic irritation, trauma, hormonal imbalances or arteriovenous malformation. Our case did not have any episodes of chronic irritation, and we could not determine the etiology. Angiosarcomas, leiomyosarcoma, hemangioendotheliomas and papillary endothelial hyperplasia should be considered in the differential diagnosis of intravenous pyogenic granuloma (IVPG).

Capillary vessels covered proliferation of the endothelial cells and fibroblast cells microscopically are characteristic of PG. The capillary cells are structured in a lobular fashion and so have recently been called lobular capillary hemangioma (LCH). Cawson et al. stated that PG can be categorized into two types histopathologically: LCH and non-LCH. LCH has lobular proliferation, a small luminal diameter and α-SMA-positive perivascular mesenchymal cells, while non-LCH has no lobular proliferation, a larger luminal diameter and α-SMA-negative perivascular mesenchymal cells. We diagnosed the present tumor as LCH-type PG [5].

IVPGs have been reported about 50 cases. Among these cases, 23 did not originate at a typical site, like the oral cavity or the upper extremities. There have been no other reports of tumors originating in the cardiac or pulmonary artery [6–11]. Among the 23 cases, IVPGs originating in the urinary organs were most frequent (17 cases). The other cases originated in the iliac vein, azygos vein, adrenal gland, subclavian vein, ovary and internal jugular vein (on each). We cannot explain why so many cases originated in the urinary organs.

Some recurrent cases have been reported, but those were considered to have been caused by incomplete resection. As IVPG is unlikely to spread through the bloodstream [11], the risk of recurrence is rare as long as complete tumor resection is accomplished. We performed this operation as lobectomy, not pneumonectomy, because there was no evidence that this

tumor was malignant according to pre-operative examinations and intra-operative findings. If the tumor had been suggested to be malignant, or if we had noted that the tumor had reached the left pulmonary lower artery branches, we would have had to perform pneumonectomy to prevent the risk of recurrence. Compared with lobectomy, pneumonectomy is more likely to be associated with cardiac or respiratory complications, such as atrial fibrillation and pulmonary hypertension [12]. After left upper lobectomy, we successfully achieved left pulmonary artery benign tumor resection and pulmonary artery plasty using a self-pericardial patch under cardiopulmonary bypass.

Conclusion

To our knowledge, this is the first case of PG originating in the pulmonary artery. As the tumor was likely to be benign, we avoided pneumonectomy when resecting this pulmonary artery tumor.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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