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CASE REPORT

Resection of a plasma cell granuloma combining a conventional posterolateral left-sided thoracotomy with a minimally invasive valve approach

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Abstract
Plasma cell granuloma (PCG) is a rare benign tumor that is difficult to differentiate from malignancy. Depending on the location of the PCG, surgical management can be challenging. We describe a patient with a PCG involving the left lower lobe extending into the left atrium, that was resected en bloc using a conventional posterolateral thoracotomy combined with a surgical approach predominantly used for minimally invasive mitral valve surgery. This case illustrates how it is possible to utilize a technique used for cardiac surgery for tumors of pulmonary origin involving the heart.

Keywords Plasma Cell Granuloma · Posterolateral thoracotomy · Minimally invasive valve surgery · Lobectomy

Introduction

Plasma cell granuloma (PCG) is considered to be a rare benign neoplastic lesion affecting younger patients [1]. The terminology around PCG is inconsistent, and in the literature the tumors are also named inflammatory pseudotumor, fibrous histiocytoma, or fibroxanthoma [2]. Depending on the location of the PCG, surgical management can be challenging. We report a case of a PCG involving the left lower lobe (LLL) extending into the left atrium. The LLL and the tumor and its extension were resected en bloc using a conventional posterolateral thoracotomy combined with a surgical approach predominantly used for minimally invasive mitral valve surgery.

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Case report

A 36-year-old woman, former smoker without previous medical history, presented with a few days of dyspnea, wheezing breathing and hemoptysis. Pulmonary X-ray showed a density in the LLL, and follow-up computer tomography confirmed the finding of a tumor measuring $9 \times 7 \times 6$ cm (Fig. 1). Both conventional bronchoscopy and endobronchial ultrasound-directed fine needle aspiration from the tumor in the superior segment of the LLL contained malignant cells. The final pathology report described a tumor that was either a sarcoma or a PCG. The patient was presented at a multidisciplinary conference, and magnetic resonance imaging was performed confirming the location of the tumor as well as the tumor extension into the left atrium. The tumor protruded through the left lower pulmonary vein into the left atrium with no radiologic sign of infiltration. It was decided to perform a lobectomy of the LLL together with the tumor component in the left atrium, en bloc.

The patient was placed in a right decubitus position with a left-sided double-lumen endotracheal tube to facilitate single lung ventilation. Approach was made via a left-sided conventional posterolateral thoracotomy below costa five. The tumor consolidated the LLL with adhesions to both the diaphragm and pericardium, but the lobe was not congested. The LLL was prepared as a for a standard lobectomy and the left lower pulmonary vein was dissected free from surrounding tissue, the pericardium opened, and the

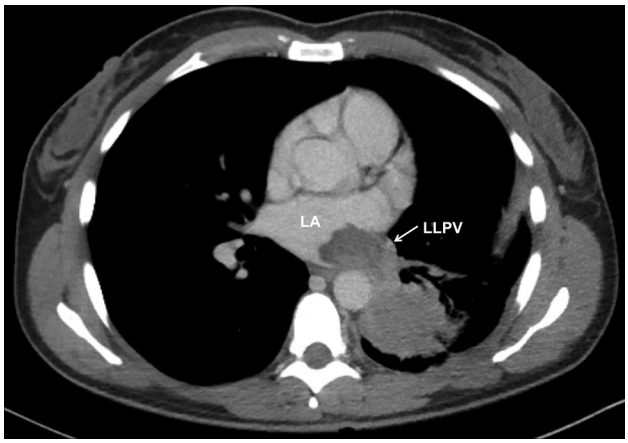

Fig. 1 Computer tomography image of the tumor in the left lower lobe extending into the left lower pulmonary vein (LLPV) and the left atrium (LA)

tumor palpated within the vein as well as the left atrium (Fig. 2a). Heparin (3 mg/kg) was given and a 25 Fr two-stage cannula (RAP 23/25, Sorin Group, Mirandola, Italy) was introduced through the left femoral vein and placed in the right atrium using transesophageal echocardiographic guidance. A 21 Fr arterial cannula (EndoReturn, Edwards Lifesciences, Irvine, CA, USA) was introduced into the left femoral artery (Fig. 2b). An intra-aortic occlusion device (IntraClude, Edwards Lifesciences, Irvine, CA, USA) was placed in the ascending aorta using transesophageal echocardiographic guidance introduced through the side-arm of the EndoReturn cannula. The occlusion device was filled with saline and asystole induced with adenosine and antegrade cold blood cardioplegia [3]. The left atrium was opened around the left lower pulmonary vein and the tumor component exposed. The tumor was found to be nonadherent to the atrial wall, and was removed en bloc with the LLL with a cuff of atrial tissue around the left lower pulmonary vein (Fig. 2c). Total aortic cross-clamping time and time on

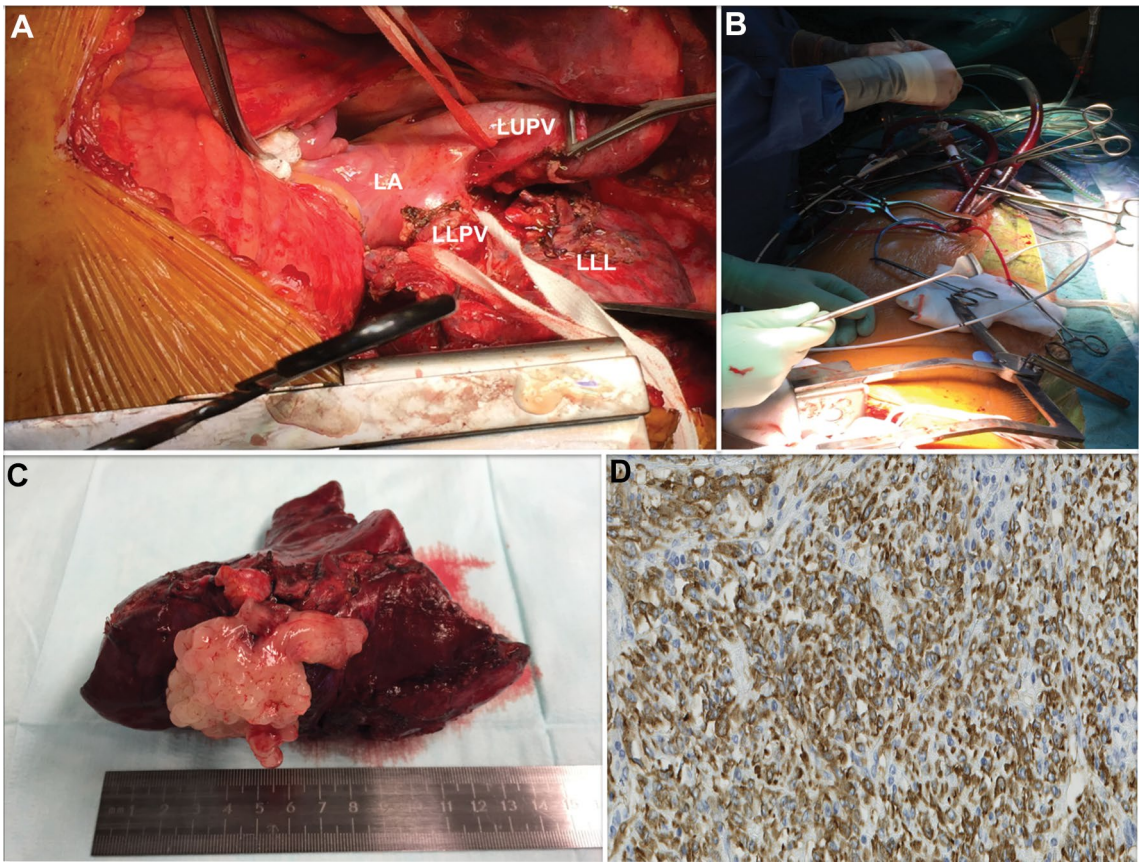

Fig. 2 Intraoperative image showing the tumor in the left lower lobe (LLL) and its relationship with the left atrium (LA). A surgical snugger was placed around the left upper and lower pulmonary vein (LUPV and LLPV) (**a**). Intraoperative image showing the peripheral cardiopulmonary bypass cannulation and the conventional posterolateral left-sided thoracotomy (**b**). The tumor in the LLL and its extension into the LA were resected en bloc (**c**). Histological section shows the atypical cells staining positive for anaplastic lymphoma kinase (**d**)

cardiopulmonary bypass were 20 and 41 min, respectively. The thoracotomy was closed after giving protamine and the patient had been weaned uneventfully from cardiopulmonary bypass. The patient recovered speedily from the procedure and was discharged home 10 days after surgery. The final pathology examination confirmed the radical resection of a PCG of unknown malignant potential as well as three negative lymph nodes. The tumor contained a large number of plasma cells (confirmed with CD138), and was positive for smooth muscle actin and vimentin. Furthermore, the tumor stained positive for anaplastic lymphoma kinase and had an elevated IgG4/IgG ratio (Fig. 2d). The resection margins were negative. At clinical follow-up 22 months after the index operation, the patient had no signs of recurrence of the tumor in the thorax.

Discussion

PCG, or inflammatory pseudotumor, is a rare lesion that can occur in almost any organ including the lung, and is predominantly intraparenchymal. Cardiac involvement, however, is rarely seen [4]. PCG can be interpreted clinically and radiologically as cancer. Imaging modalities such as computer tomography and magnetic resonance imaging are essential to define the location and the extent of the PCG. PCG is difficult to differentiate from a malignancy and fine needle aspiration or histologic frozen sections are often inconclusive [2]. Furthermore, complete resection of the PCG is essential to reduce the risk for recurrence. A PCG is often associated with pathologic IgG4 levels, and an elevated serum concentration of IgG4 may help in the differentiation between PCG and other tumors. The existence of circulating plasmablasts may be even more sensitive as a marker than raised IgG4 levels when diagnosing IgG4-releated disease [5], but this was not investigated in our patient. An elevated ratio between IgG4 and IgG has also been proposed as a specific marker of PCG [5], as illustrated in our case.

The minimally invasive mitral valve surgery approach used in this case report, is usually done from the right side. In terms of cannulation, de-airing of the heart using a left-side approach did not pose any unique procedural challenges. The use of the intra-aortic occlusion devise (placed in the ascending aorta using the side-arm of the EndoReturn cannula) facilitates aortic cross-clamping from the left side, since using a conventional cross-clamp can occasionally be troublesome from this side. Contraindications to the procedure are same as with the traditional use of the surgical platform for minimally invasive mitral valve surgery; signs of peripheral vascular disease, major aortic regurgitation and dilated aorta ascendens. If any of these contraindications are present, the proposed surgical approach cannot be used. The wide open surgical field used in this case ensures a safe operative field, despite the fact that a left-sided thoracotomy is rarely used for intracardiac procedures.

In this case report we describe a patient with a PCG located in the LLL and extending into the left atrium, posing a surgical challenge. The tumor could be resected en bloc via a left-sided conventional posterolateral thoracotomy with the surgical platform normally used at our department to perform minimally invasive mitral valve surgery. This particular case illustrates the fact that novel surgical techniques and methodologies, such as the approach used to perform minimally invasive cardiac valve surgery via a thoracotomy, can be expanded to complement traditional surgical procedures in the treatment of other non-cardiac diseases.

Compliance with ethical standards

Conflict of interest The authors have declared that no conflict of interest exists.

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