Reconstruction surgery case report: ex vivo surgery and auto lung transplantation for pulmonary artery sarcoma

Daisuke Nakajima, Sou Miyahara, Hiroshi Date

Department of Thoracic Surgery, Kyoto University, Kyoto, Japan

Correspondence to: Hiroshi Date, MD, PhD. Department of Thoracic Surgery, Kyoto University, 54 Shogoin-kawahara-cho, Sakyo-ku, Kyoto 606-8507, Japan. Email: hdate@kuhp.kyoto-u.ac.jp.

Abstract: This is a report on the lung autotransplant techniques for the treatment of intimal sarcoma of pulmonary artery (PA). A 58-year-old man was diagnosed with PA sarcoma occluding the right main PA and peripherally extending into the right interlobar PA. First, the tumor was completely removed by pulmonary endarterectomy combined with right pneumonectomy under cardiopulmonary bypass. A following ex vivo evaluation of the explanted right lung revealed that the right lower PA was intraluminally intact, so the right lower lobe was used for reimplantation after being flushed with cold organ preservation solution on a back table. The patient was safely weaned from cardiopulmonary bypass following transplantation. His early post-transplant course was uneventful. He died of tumor recurrence at the distal left PA and liver and bone metastasis 14 months after surgery.

Keywords: Lung autotransplantation; pneumonectomy; pulmonary artery sarcoma; pulmonary endarterectomy; case report

Received: 27 September 2019; Accepted: 22 October 2019; Published: 06 January 2020.

doi: 10.21037/jovs.2019.10.06

View this article at: http://dx.doi.org/10.21037/jovs.2019.10.06

Introduction

Pulmonary artery (PA) sarcoma has been reported as an extremely rare tumor with a very poor prognosis. Surgical intervention, including pulmonary endarterectomy and pneumonectomy, is considered the mainstay of treatment to relieve symptoms and offer a chance for a long-term survival (1-5). However, the optimal surgical approach must be determined individually, based on the tumor location and the patients’ clinical condition.

Lung autotransplantation has been successfully employed for various types of lung disease in order to spare the pulmonary parenchyma, preserving the lung function (6-15). We herein report our experience performing lung autotransplantation with ex vivo bench surgery for the treatment of rare intimal sarcoma of the PA in accordance with the CAse REport (CARE) guideline (16).
of predicted value). The patient’s condition rapidly deteriorated and he became bed-bound. Based on these findings, PA sarcoma was clinically suspected, and the initial surgical strategy consisted of pulmonary endarterectomy and right pneumonectomy, followed by reimplantation of the right lower lobe in order to preserve the post-operative pulmonary function.

The surgical procedure is demonstrated in Figure 4. Through median sternotomy combined with a fourth intercostal right thoracotomy, cardiopulmonary bypass was established through the ascending aortic and bicaval cannulation. The aorta was cross-clamped, and cardiac arrest was commenced. The longitudinal incision of the PA trunk and right main PA was performed in order to remove the centrally located tumor, originating in the posterior wall of the PA immediately above the pulmonary valve. After the

**Figure 1** Enhanced chest computed tomography showed the low-attenuation filling defects occupying the entire luminal diameter of the right main pulmonary artery (PA) (A), peripherally extending into the right interlobar PA (B).

**Figure 2** Fluorodeoxyglucose-positron emission tomography/computed tomography (FDG PET-CT) demonstrated an intense uptake [maximum standardized uptake value (SUVmax) =15.2] in the intraluminal defects within the right main pulmonary artery.

**Figure 3** A pulmonary angiogram showed the right main pulmonary artery to be occluded by the intimal sarcoma.

**Figure 4** Lung autotransplant technique with ex vivo surgery for pulmonary artery sarcoma (17). Available online: http://www.asvide.com/watch/33007
right main PA had been reconstructed, the heartbeat was resumed. The cardiac arrest time was 66 minutes.

Right pneumonectomy was then performed in order to remove the residual tumor within the peripheral PA. A subsequent *ex vivo* assessment of the explanted right lung revealed that the right lower PA was intraluminally tumor-free, so the right lower lobe could be reimplanted. The right upper and middle lobes were resected *ex vivo*, and antegrade and retrograde flushing of the remaining healthy lower lobe was then performed with approximately 2,000 mL of ET Kyoto solution (Otsuka Pharmaceutical Factory, Tokushima, Japan) at 4 °C on a back table, based on our established protocol for living donor lobar lung transplantation (18,19).

We created a homologous pulmonary arterial graft conduit using a chest tube as a mould which was used for elongation of the graft PA. The implantation was performed as follows: the graft bronchus was anastomosed to the right main bronchus with running 4-0 PDS sutures for the membranous portion and interrupted 4-0 PDS sutures for the cartilaginous portion. The bronchial anastomotic site was then wrapped with a pedicled pericardial fat pad. The reconstructed graft PA was directly anastomosed to the right main PA with running 6-0 Prolene sutures. The pulmonary vein of the graft was anastomosed to the remaining upper pulmonary venous stump with running 6-0 Prolene sutures. The total ischemic time of the graft was 118 minutes, and no ischemia-reperfusion injury was noted after transplantation. The patient was successfully weaned from the cardiopulmonary support immediately after reperfusion. The cardiopulmonary bypass time was 253 minutes.

The histopathological features of the tumor were those of a pleomorphic malignancy comprising spindled and epithelioid cells admixed with inflammatory cells (*Figure 5*). Areas of necrosis and mitoses were sporadically observed. There was no local invasion of the tumor cells outside the vessel wall. These findings were consistent with a diagnosis of PA intimal sarcoma.

His early post-transplant course was uneventful: The patient was weaned from the mechanical ventilatory support on postoperative day (POD) 2 and discharged from the intensive-care unit on POD 6. He returned to his normal life one month after surgery. He died of tumor recurrence at the distal left PA and liver and bone metastasis 14 months after surgery.

**Discussion**

The lung autotransplant procedure has been previously reported as a useful technique for the treatment of various types of lung disease, including centrally located lung cancer, postpneumonectomy-like syndrome, locally advanced lung cancer, and bronchopleural fistula after right upper bronchial sleeve lobectomy (6-15). We herein reported the utility of *ex vivo* bench surgery and autotransplantation to completely resect the intimal sarcoma of the PA, thereby preserving the post-operative pulmonary function.

In the present case, the benefits of *ex vivo* surgery were considered to be as follows: (I) extended resection of tumor could be safely performed on a back table with a favorable visual field and relatively little tumor manipulation; (II) antegrade and retrograde flushes of the lung graft with cold organ preservation solution helped avoid thrombus formation in the graft and ameliorate ischemia-reperfusion-induced lung injury after reimplantation; and (III) cold lung preservation provided enough time to pathologically

*Figure 5* Histopathologic examinations of the tumor with hematoxylin and cosin staining showed atypical polymorphic spindle cells admixed with inflammatory cells. Original magnification ×20 and ×100.
confirm the tumor-free surgical margins.

PA sarcoma has been reported as an extremely rare tumor with a very poor prognosis. Although only surgical resection, commonly including pneumonectomy or pulmonary endarterectomy, is considered the mainstay of treatment to prolong the survival for patients with this orphan disease, the reported median survival time was 1.5 months without surgery and 10 to 18 months even with surgical resection (1-5). In this case, the patient died of recurrence of PA sarcoma 14 months postoperatively. However, no major complications were observed in the early postoperative course, and he was able to return to his normal life one month after surgery, although he was completely bed-bound before surgery.

Conclusions
We reported a case of PA intimal sarcoma that was completely resected with pulmonary endarterectomy and right pneumonectomy, followed by ex vivo surgery and lung autotransplantation. This complicated procedure was found to be technically feasible without any major post-operative complications and may be an optimal surgical approach for select patients with PA sarcoma.

Acknowledgments
None.

Footnote
Conflicts of Interest: The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. Informed consent was obtained from the patient for publication of this manuscript and any accompanying images.

References
17. Nakajima D, Miyahara S, Date H. Lung autotransplant...


doi: 10.21037/jovs.2019.10.06