Successful Infant Pneumonectomy with Unilateral Pulmonary Artery Occlusion Test

Koji Kato, MD, Taichi Kato, MD, Satoshi Hayano, MD, Yoshie Fukasawa, MD, Atsushi Numaguchi, MD, Tetsuo Hattori, MD, Akiko Saito, MD, Yoshiaki Sato, MD and Masahiro Hayakawa, MD

Summary

The use of unilateral pulmonary artery occlusion (UPAO) test for the preoperative evaluation of pneumonectomy was reported in adult patients. On the contrary, in infants, no strategies have yet been recommended to predict hemodynamics after pneumonectomy, nor has use of the UPAO test been reported. We describe the first case of infant with abnormal pulmonary circulation in whom successful pneumonectomy was performed after preoperative evaluation using UPAO test. Right pneumonectomy was planned for an 8-month-old girl, because of decreased right pulmonary function, high risk of pneumothorax, and impaired left lung expansion due to overexpansion caused by severe left bronchial stenosis and bronchomalacia. However, she had also prolonged pulmonary hypertension and there was difficulty in accurate echocardiographic evaluation of its severity due to concomitant left pulmonary artery stenosis. Furthermore, contrast-enhanced computer tomography suggested a certain degree of right pulmonary venous flow, discordant with the result showing scarce right pulmonary flow in perfusion scintigraphy. Predicting postoperative hemodynamic changes was therefore considered difficult. To evaluate these concerns, we performed cardiac catheterization and UPAO test to simulate postoperative hemodynamics. Pulmonary arteriography showed decreased but significant right pulmonary arterial and venous flows. Measurements including pulmonary artery pressure and cardiac index showed no marked changes after occlusion. Based on UPAO test results, the operation was successfully performed and hemodynamics remained stable postoperatively. The UPAO test may be useful for infants with cardiopulmonary impairment to evaluate the tolerability of pneumonectomy.

Key words: Preoperative evaluation, Pulmonary hypertension, Pulmonary artery stenosis

Pneumonectomy is rarely performed for children, but it is the principal treatment for severe congenital lung malformations, including congenital pulmonary airway malformation (CPAM). Postoperative decreases in lung volume lead to reduced ventilation and a decrease in the pulmonary vascular bed, resulting in an increased risk of pulmonary hypertension and right heart failure. For adult patients, several preoperative assessments have been used to predict postoperative complications for lung resection, including both lung function tests and imaging tests. In addition, the unilateral pulmonary artery occlusion (UPAO) test is performed to assess the safety of surgery for patients considered to have reduced cardiopulmonary function and simulates a decrease in the pulmonary vascular bed after pneumonectomy. On the contrary, because lung function tests are not applicable for infants, preoperative evaluations rely solely on imaging tests. No strategies have yet been recommended for predicting complications affecting cardiopulmonary function after pneumonectomy in infants, nor has use of the UPAO test been described. We present the case of an infant for whom postoperative complications were a concern because of prolonged pulmonary hypertension, and in whom pneumonectomy was safely performed after preoperative evaluation using the UPAO test.

Case Report

A female infant was born at 37 weeks of gestation by emergent cesarean section due to fetal distress. Large right CPAM had been suspected at 18 weeks of gestation from fetal ultrasonography and magnetic resonance imaging.

At delivery, the patient was cyanotic and hypotonic, and we performed initial resuscitation and intubation. Apgar scores were 4 at 1 minute and 5 at 5 minutes, and birth weight was 1884 g. Radiography showed a large mass in the right chest. Echocardiography revealed a flat-
tended intraventricular septum on the left ventricular short-axis view and right-to-left shunt flow in the ductus arteriosus, indicating severe pulmonary hypertension. We therefore administered continuous sedation and used transient inhaled nitric oxide, followed by sildenafil and beraprost.

Although the size of right lung mass gradually decreased during the neonatal period, emphysema and over-expansion of the right lung deteriorated due to a check-valve mechanism resulting from severe right bronchial stenosis and bronchomalacia. As the patient was positive end-expiratory pressure-dependent due to severe bronchomalacia, tracheotomy was performed at 5 months of age. However, pulmonary hypertension was aggravated by withdrawal of continuous sedation after tracheotomy. We reinstituted continuous sedation and also administered bosentan, as well as beraprost and sildenafil. In addition, pulmonary ventilation-perfusion scintigraphy showed progressive right pulmonary dysfunction. At 8 months of age, right/left pulmonary function ratios were 3/97 in ventilation and 2/98 in perfusion scintigraphy, compared with 23/77 and 36/64 at 3 months of age, respectively. Three-dimensional computed tomography (CT) at 1 month of age showed hypoplasia of the left pulmonary artery (Figure 1A), and this was improved only at the left distal pulmonary artery, resulting in regional left peripheral pulmonary artery stenosis at 8 months of age (Figure 1B).

We considered right pneumonectomy because of impaired left lung expansion, absence of right pulmonary respiratory function, and the risk of pneumothorax due to overexpansion of the right lung. However, the left pulmonary artery stenosis made the degree of pulmonary hypertension difficult to accurately evaluate. Furthermore, CT at 8 months of age suggested a certain degree of right pulmonary venous flow, discordant with the results of scintigraphy. Predicting postoperative hemodynamic changes was therefore considered difficult. To evaluate these concerns, we performed cardiac catheterization. Pulmonary arteriography showed decreased but significant right pulmonary arterial (Figure 2A) and right pulmonary venous flows. Aortic angiography showed no collateral vessels to the right lung mass. We therefore performed the UPAO test (Figure 2B) to simulate the hemodynamic status after right pneumonectomy. Before and after occlusion for 5 minutes, mean ratios of left peripheral pulmonary artery pressure to systemic artery pressure (Pp/Ps) were 39/83 and 38/82, respectively, and systolic Pp/Ps were 48/114 and 47/119, respectively. Similarly, other indices were constant before and after occlusion, with total pulmonary vascular resistance index (TPVRI) at 488 dyne·s/cm$^2$/m$^2$ and 481 dyne·s/cm$^2$/m$^2$ and cardiac index (CI) at 6.39 L/minute/m$^2$ and 6.32 L/minute/m$^2$, respectively. According to these results of stable hemodynamics under the UPAO test, we judged that the patient would tolerate the postoperative hemodynamic changes.

Hemodynamics after right pneumonectomy at 8 months of age remained stable. At 14 months of age, the patient underwent external tracheobronchial stabilization for tracheo- and left bronchomalacia. As of the time of writing, the patient is 2 years of age and free of mechanical ventilation during waking hours. Although we did not conduct cardiac catheterization after the pneumonectomy, right ventricular systolic function using tricuspid annular plane systolic excursion did not change after the pneumonectomy and no symptoms or signs of right heart failure have been encountered.

**Discussion**

This represents the first case report to suggest the usefulness of the UPAO test for infants. Pneumonectomy carries a risk of significant pulmonary hypertension and even a fatal course in infants. Our case involved difficulty predicting postoperative hemodynamics, including pulmonary hypertension. We thus conducted cardiac catheterization followed by the UPAO test to evaluate pulmonary hypertension and simulate the postoperative hemodynamic state.

The following operative indications from the UPAO test for pneumonectomy concerning pulmonary circulation...
have been proposed for adulthood:\(^6\): TPVRI < 700 dyne·s/cm\(^2\)/m\(^2\); mean pulmonary artery pressure (PAP) < 30 mmHg; and CI > 2.0 L/minute/m\(^2\). However, no criteria have been proposed for infants. Our case revealed that TPVRI and CI during occlusion were within the proposed limits, whereas mean PAP was higher than the suggested criterion. However, we considered that the patient would tolerate the operation, because the measured parameters showed no changes after occlusion, and hemodynamics remained stable after pneumonectomy. The UPAO tests for adult patients were performed for 20-30 minutes in previous reports.\(^7\) On the contrary, we occluded for 5 minutes because we intended to avoid prolonged anesthesia. Furthermore, measured parameters are likely to change in a few minutes if certain amount of right pulmonary arterial flow exists. However, adequate occlusion time in the UPAO test is also a subject of future investigation.

In conclusion, the UPAO test appears beneficial as a preoperative investigation to evaluate the tolerability of pneumonectomy for infants showing cardiopulmonary dysfunction. Further experience and accumulation of data are needed to ensure the efficacy of the UPAO test and define appropriate criteria for the safe management of pneumonectomy in infants.

Disclosures

Conflicts of interest: None.

References

1. Harpole DH Jr, DeCamp MM Jr, Daley J, et al. Prognostic models of thirty-day mortality and morbidity after major pulmonary resection. J Thorac Cardiovasc Surg 1999; 117: 969-79.
2. Nagamatsu Y, Shima I, Hayashi A, Yamana H, Shirouzu K, Ishitake T. Preoperative spirometry versus expired gas analysis during exercise testing as predictors of cardiopulmonary complications after lung resection. Surg Today 2004; 34: 107-10.
3. Nagamatsu Y, Ohkita A, Kimura NY, et al. Retrospective investigation of pulmonary resection in patients with high total pulmonary vascular resistance during preoperative unilateral pulmonary artery occlusion. Gen Thorac Cardiovasc Surg 2009; 57: 244-9.
4. Ben-Ishay O, Nicksa GA, Wilson JM, Buchmiller TL. Management of giant congenital pulmonary airway malformations requiring pneumonectomy. Ann Thorac Surg 2012; 94: 1073-8.
5. Schwartz MZ, Ramachandran P. Congenital malformations of the lung and mediastinum- a quarter century of experience from a single institution. J Pediatr Surg 1997; 32: 44-7.
6. Tanita T, Tomoyasu M, Deguchi H, Mizuno M, Kawazoe K. Review of preoperative functional evaluation for lung resection using the right ventricular hemodynamic functions. Ann Thorac Surg 2004; 10: 333-9.
7. Ikeda T, Onuki T, Nishiuichi M, Kanzaki M, Nitta S. Clinical assessment of unilateral pulmonary artery occlusion test with dobutamine infusion. Expansion of surgical indications in patients with primary lung cancer and impaired cardiopulmonary functions. Jpn J Thorac Cardiovasc Surg 2000; 48: 802-8.