

Cite this article as: Scholl H, Lutz JA, Kadner A, Schmid RA. Dynamic stenosis of the right main bronchus in a 3-month-old child: a tracheobronchial sleeve resection under venovenous extracorporeal membrane oxygenation. *Eur J Cardiothorac Surg* 2018; doi:10.1093/ejcts/ezy256.

Dynamic stenosis of the right main bronchus in a 3-month-old child: a tracheobronchial sleeve resection under venovenous extracorporeal membrane oxygenation

Heiko Scholl^a, Jon A. Lutz^{a,*}, Alexander Kadner^b and Ralph A. Schmid^a

^a Division of General Thoracic Surgery, Inselspital, Bern University Hospital, University of Bern, Bern, Switzerland

^b Department for Cardiovascular Surgery, Centre for Congenital Heart Disease, Inselspital, Bern University Hospital, University of Bern, Bern, Switzerland

* Corresponding author. Division of General Thoracic Surgery, Inselspital, Bern University Hospital, University of Bern, 3010 Bern, Switzerland.
Tel: +41-31-6327567; fax: +41-31-6322327; e-mail: jon.lutz@insel.ch (J.A. Lutz).

Received 8 May 2018; received in revised form 18 June 2018; accepted 20 June 2018

Abstract

We report the case of a boy with congenital dynamic stenosis of the right main bronchus. The operation was postponed to a later date, when the patient was in a better clinical position to tolerate surgery. A tracheobronchial sleeve resection under dual-lumen venovenous extracorporeal membrane oxygenation was performed.

Keywords: Congenital tracheobronchial stenosis • Tracheobronchial sleeve-resection • Extracorporeal membrane oxygenation • Paediatric

INTRODUCTION

Congenital tracheobronchial stenosis is rare in children and usually becomes symptomatic in early adulthood [1]. This is a challenging condition in neonates in terms of perioperative airway management and the surgical repair technique. In this case, we performed a lung-sparing tracheobronchial sleeve resection under dual-lumen venovenous extracorporeal membrane oxygenation (vvECMO).

CASE

A boy was referred to us 32 days post-partum because of acute stridor and tachydyspnoea. Vaginal birth occurred at 40 weeks (weight 3445 g, length 50 cm, head circumference 33 cm and APGAR score 8/8/9). A chest X-ray showed a hyperinflated right lung with mediastinal shift (Fig. 1A). Bronchoscopy revealed dynamic stenosis of the proximal right main bronchus with the absence of 2 cartilage rings (Video 1A). The collapse of the pars membranacea produced a check valve mechanism resulting in air trapping. Echocardiography showed no heart abnormality. In the absence of cyanosis, ambulant salbutamol therapy was initiated. Discussion by the board for complex airway pathologies followed. We decided to postpone surgical treatment to after the age of 6 months. However, the boy experienced repeated and progressive cyanotic attacks; therefore, a decision for immediate surgical intervention was made.

At the time of surgery, the weight and length of the patient was 6.1 kg, and 60 cm, respectively. General anaesthesia was

initiated, and vvECMO (Avalon Elite[®] bicaval dual-lumen catheter, 13 Fr) was installed under echocardiographic guidance through the right jugular vein (Fig. 2). Oxygenation support was 186 min. The median flow was 0.39 l/min (range 0.2–0.62 l/min) with a targeted mean arterial pressure of 40 mmHg (22–58 mmHg). Through a right thoracotomy, a segmental resection of the proximal right main bronchus was performed under additional guidance by bronchoscopy. The tracheobronchial anastomosis was performed with interrupted sutures. The pars membranacea was additionally suspended posteriorly to the spine. Intraoperative bronchoscopy confirmed a patent anastomosis. Weaning from vvECMO occurred before transfer of the patient to the paediatric intensive care unit. Extubation followed on postoperative day 1 after control bronchoscopy (Video 1B). A postoperative chest X-ray showed a reduction in mediastinal shift (Fig. 1B).

On postoperative day 3, the boy presented with seizures on the left side, with ictal signs on the right side of the EEG. Magnetic resonance imaging (MRI) showed signs of acute ischaemia of the right hemisphere. Symptoms, however, resolved spontaneously, and the boy was discharged on postoperative day 13. At the time of the last follow-up, 11 months after surgery, the patient was well.

DISCUSSION

Postponing the operation in this child allowed 2 months of additional growth with a weight gain of 2.5 kg despite recurrent hospitalizations. This strategy, in an initially non-respiratory compromised patient, made the operation easier for both surgical and anaesthesiology teams.

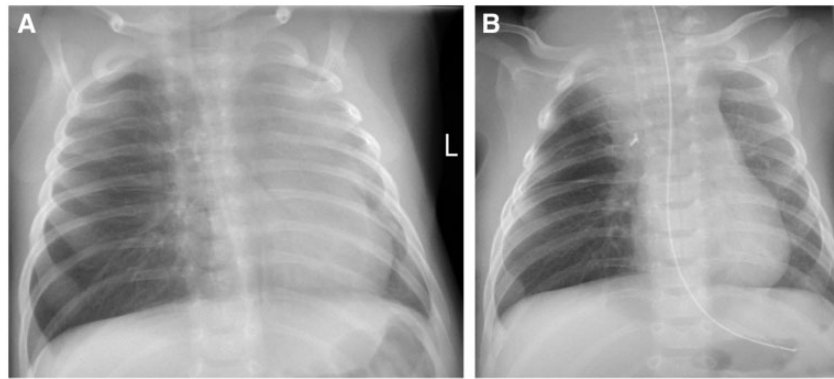


Figure 1: (A) A preoperative chest X-ray showing the hyperinflated right lung and mediastinal shift to the left. (B) A postoperative (Day 3) chest X-ray showing a reduction in mediastinal shift. Haemostatic clip in projection of the right hilum.



Video 1: (A) Preoperative bronchoscopy showing the narrowing and the check valve mechanism at the tracheobronchial angle. (B) Postoperative bronchoscopy showing the patent and stable anastomosis.

There are different approaches to treating a stenotic airway segment [1, 2]. Bronchoscopy with balloon dilatation and/or stenting is seldom a definitive treatment and is linked to major complications and reinterventions. The localization of the stenosis at the tracheobronchial angle would have made a carinal stent necessary, putting the whole airway at risk in case of mucous plugging, formation of granulation tissue or stent dislocation. Slide tracheoplasty was not suitable because of the shortness of the stenosis. Therefore, we choose to perform a tracheobronchial sleeve resection and an anastomosis with posterior tension sutures to help maintain the patency of the right main bronchus.

The main challenges for airway surgery in young patients are safety of anaesthesia and operative field visibility. vVECMO offers an unimpeded view and avoids the need for ventilation tubes in the surgical field. Single-vessel cannulation with a bicaval dual-lumen catheter has been described in the paediatric population [3] and is routinely used at our institution [4]. The neurovascular complication that occurred is well known in the long-term extracorporeal membrane oxygenation support for cardiorespiratory failure [5]. It is not clear whether the cannula located in the internal jugular vein compressed the right main carotid artery, whether ischaemia was due to intermittent periods of hypotension during surgery or whether the reason was a combination of both. Fortunately, the patient recovered without any impairment.



Figure 2: Photograph showing the inserted dual-lumen venovenous extracorporeal membrane oxygenation.

CONCLUSION

The timing of the surgery and the type of surgery are crucial in young patients with airway stenosis and should be discussed by a

specialized interdisciplinary team. vvECMO offers a continuous oxygenation support during intervention and allows an optimal access to the surgical field. Careful assessment for potential intra-operative episodes of cerebral hypoperfusion or hypoxia while on vvECMO support is mandatory.

ACKNOWLEDGEMENTS

The authors thank Gregor J. Kocher and Carmen Casaulta for their implications in the management of the case and relecture of the manuscript.

Conflict of interest: none declared.

REFERENCES

- [1] Poupalou A, Varetti C, Lauron G, Steyaert H, Valla JS. Perinatal diagnosis and management of congenital bronchial stenosis or atresia: 4 cases. *J Thorac Cardiovasc Surg* 2011;141:e11–4.
- [2] Hewitt RJ, Butler CR, Maughan EF, Elliott MJ. Congenital tracheobronchial stenosis. *Semin Pediatr Surg* 2016;25:144–9.
- [3] Fallon SC, Shekerdeman LS, Olutoye OO, Cass DL, Zamora JJ, Nguyen T *et al.* Initial experience with single-vessel cannulation for venovenous extracorporeal membrane oxygenation in pediatric respiratory failure. *Pediatr Crit Care Med* 2013;14:366.
- [4] Kocher GJ, Zehnder A, Erdoes G, Seidl C, Winkler B, Schmid RA. Single-cannula, single-incision thoracoscopic anatomic segmentectomy after pneumonectomy. *J Thorac Cardiovasc Surg* 2017;154:e29–31.
- [5] Hervey-Jumper SL, Annich GM, Yancon AR, Garton HJL, Muraszko KM, Maher CO. Neurological complications of extracorporeal membrane oxygenation in children. *J Neurosurg Pediatrics* 2011;7:338–44.