Can J Anesth/J Can Anesth (2022) 69:265–268
https://doi.org/10.1007/s12630-021-02148-2

CASE REPORTS / CASE SERIES

Acute plastic bronchitis after Ross procedure treated with veno-venous extracorporeal membrane oxygenation

Bronchite plastique aiguë après procédure de Ross traitée par oxygénation par membrane extracorporelle veino-veineuse

Charles St-Arnaud, MD · Mohamed Abdel Halim, MD · Michael Mayette, MD

Received: 18 August 2021 / Revised: 3 October 2021 / Accepted: 4 October 2021 / Published online: 2 December 2021
© Canadian Anesthesiologists' Society 2021

Abstract

Purpose *Plastic bronchitis is a rare condition characterized by the formation of airway casts occluding the lower respiratory tract. It is described more commonly in children, especially following correction of congenital heart disease. It involves lymphatic abnormalities leading to endobronchial lymph precipitating airway cast formation. When it presents acutely, it can lead to acute airway obstruction, which can be life-threatening. Plastic bronchitis has been rarely described in adults and is potentially underdiagnosed. The purpose of this case report is to emphasize, for the adult anesthesiologist and adult critical care physician, the importance of prompt diagnosis and respiratory support in a case of plastic bronchitis.*

Clinical features *A 40-yr-old female with severe aortic stenosis underwent a Ross procedure. The surgery was uneventful, but within two hours of arrival in the intensive care unit, the patient developed severe hypoxemia. Despite attempts to optimize her respiratory status, the patient remained severely hypoxemic, and veno-venous extracorporeal membrane oxygenation (ECMO) was initiated using a percutaneous femoro-femoral cannulation. A bronchoscopy showed bronchial secretions casting the proximal bronchus, suggestive of plastic bronchitis. After numerous bronchoscopies, we were able to clean the airways and wean the ECMO support on postoperative day 3.*

Conclusion *Plastic bronchitis can present in adult patients and be life-threatening when associated with acute respiratory failure. We report an unusual case of an adult patient treated with veno-venous ECMO for plastic bronchitis following cardiac surgery. Use of ECMO support while providing airway cleaning can be lifesaving in patients with respiratory failure secondary to plastic bronchitis.*

Résumé

Objectif *La bronchite plastique est une affection rare caractérisée par la formation de bouchons muqueux qui moulent et obstruent les voies aériennes inférieures. Elle est plus fréquemment décrite chez les enfants, en particulier après la correction d'une cardiopathie congénitale. Elle découle d'anomalies lymphatiques conduisant à l'accumulation de lymphe endobronchique, précipitant la formation de bouchons muqueux dans les voies aériennes. Lorsqu'elle se présente de manière aiguë, la bronchite plastique peut entraîner une obstruction aiguë des voies aériennes, une complication potentiellement fatale. La bronchite plastique a rarement été décrite chez l'adulte et est potentiellement sous-diagnostiquée. L'objectif de cette présentation de cas est de souligner, pour l'anesthésiologiste et l'intensiviste s'occupant d'une population adulte, l'importance d'un diagnostic rapide et d'un support respiratoire en cas de bronchite plastique.*

Caractéristiques cliniques *Une femme de 40 ans souffrant d'une sténose aortique sévère a bénéficié d'une procédure de Ross. La chirurgie s'est déroulée sans*

C. St-Arnaud, MD (✉)
Department of Medicine, Internal Medicine and Critical Care Division, Université de Sherbrooke, 3001, 12e Avenue Nord, Sherbrooke, QC J1H 5N4, Canada
e-mail: Charles.st-arnaud@usherbrooke.ca

M. A. Halim, MD
Department of Surgery, Cardiac Surgery Division, Centre Hospitalier Universitaire de Sherbrooke, Université de Sherbrooke, Sherbrooke, QC, Canada

M. Mayette, MD
Department of Medicine, Internal Medicine and Critical Care Division, Centre Hospitalier Universitaire de Sherbrooke, Université de Sherbrooke, Sherbrooke, QC, Canada

incident, mais dans les deux heures suivant son arrivée à l'unité de soins intensifs, la patiente a présenté une hypoxémie sévère. Malgré les tentatives d'optimisation de son état respiratoire, la patiente est restée gravement hypoxémique et une oxygénation par membrane extracorporelle (ECMO) veino-veineuse a été amorcée à l'aide d'une canulation fémoro-fémorale percutanée. Une bronchoscopie a montré des sécrétions bronchiques moulant les bronches proximales, évoquant une bronchite plastique. Après de nombreuses bronchoscopies, nous avons pu nettoyer les voies aériennes et sevrer la patiente du soutien ECMO au 3ème jour postopératoire.

Conclusion *La bronchite plastique peut se présenter chez les patients adultes et être potentiellement fatale lorsqu'elle est associée à une insuffisance respiratoire aiguë. Nous rapportons un cas inhabituel d'une patiente adulte traitée par ECMO veino-veineuse pour une bronchite plastique après une chirurgie cardiaque. L'utilisation du soutien par ECMO simultanément au nettoyage des voies aériennes peut être nécessaire chez les patients atteints d'insuffisance respiratoire secondaire à une bronchite plastique.*

Keywords ross · plastic bronchitis · ECMO

Plastic bronchitis is characterized by bronchial obstruction by endobronchial plugs of rubber-like consistency and is rarely associated with cardiac surgery. It presents with signs of respiratory distress related to acute airway obstruction by airway casts. Severity of symptoms upon presentation depends on the severity of airway obstruction. In severe cases, the challenge is to clear the airways while providing adequate respiratory support. We report an unusual case of plastic bronchitis complicating cardiac surgery treated with extracorporeal membrane oxygenation (ECMO) to allow bronchial cleaning until restoration of lung function.

Case report

A 40-yr-old female patient presented with a bicuspid aortic valve and severe aortic stenosis. She had no significant comorbidities. She underwent a Ross procedure because of her young age and favorable anatomy. The aortic valve was replaced with the patient's pulmonary valve (autograft), and the pulmonary valve was replaced by a homograft (CryoValve SG pulmonary human heart valve; CryoLife, Inc., Kennesaw, GA, USA). The surgery was uneventful, and the patient was transferred to the surgical intensive care unit in stable condition.

Within two hours of arrival in the intensive care unit, the patient developed severe hypoxemia. Despite attempts to optimize her respiratory status using high levels of sedation, paralytic agents, low tidal volumes, high positive end-expiratory pressure, and lung recruitment, the patient remained severely hypoxemic with arterial oxygen partial pressure/fraction of inspired oxygen (PaO_2/F_iO_2) ratio of 75. Transesophageal echocardiography showed normal right and left heart function and a competent neo-aortic valve and pulmonary homograft. Our investigations included a chest *x-ray*, which showed diffuse opacity of the right lower lung, and bronchoscopy, which showed thick secretions all over the lower respiratory tract. A computed tomography scan of the chest (Figure 1, panel A) performed six hours later showed complete consolidation of the right lung and left lower lobe.

Eight hours following admission, despite maximal respiratory and hemodynamic support, the patient developed a low-flow state with multi-organ failure. Because of the underlying respiratory condition suggestive of fulminant acute respiratory distress syndrome, veno-venous ECMO was initiated using a percutaneous femoro-femoral cannulation with a 25-Fr access canula in the right femoral vein and a 21-Fr return canula in the left femoral vein with its distal tip at the junction of the inferior vena cava and the right atrium. Upon initiation of the ECMO support, the patient's condition stabilized with instantaneous normalization of the blood gas and restoration of hemodynamic stability.

A bronchoscopy repeated on postoperative day 1 showed bronchial secretions casting the right main stem bronchus and the left lower lobe with extension as a "saddle bronchio-tracheal cast" up to the distal trachea (Figure 1, panel B). Microscopic examination of a bronchial cast (Figure 1, panel C) revealed hypocellular material containing layers of mucin and fibrin with occasional epithelial debris cells, compatible with plastic bronchitis. Between postoperative days 1 and 3, numerous flexible bronchoscopies were performed to remove bronchial casts using normal saline lavage and biopsy forceps. N-acetylcysteine 20% was nebulized every six hours. Dornase alfa, a deoxyribonuclease enzyme used as a mucolytic agent in cystic fibrosis, was instilled in all affected pulmonary segments during a bronchoscopy performed on postoperative day 2. Extracorporeal membrane oxygenation was weaned on postoperative day 3, and the patient was extubated on postoperative day 4. Following extubation, the hospital course was uneventful and the patient was discharged home on postoperative day 14. Upon 18-month follow-up, she was doing well and showed no sign of active lung disease.

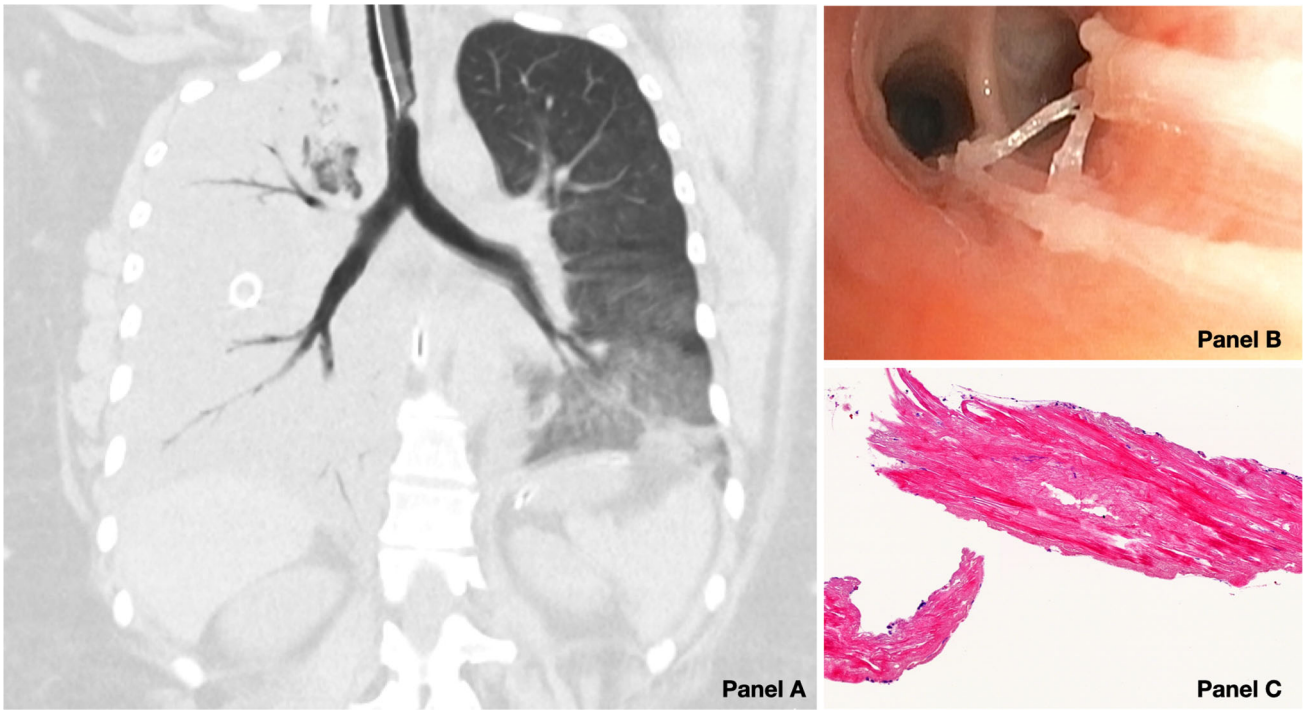

Figure 1 Panel A: Computed tomography scan of the chest (coronal view) performed six hours following admission to the intensive care unit showed complete right lung and left lower lobe consolidation. Panel B: Bronchial cast extracted with a flexible bronchoscope. Panel C: Hematoxylin and eosin staining with 200× scale of a bronchial cast showing hypocellular material containing layers of mucin and fibrin with occasional amorphous epithelial debris cells. Absence of muscle cells or inflammatory infiltrate was confirmed with immunohistochemical staining.

Discussion

Plastic bronchitis is characterized by endobronchial plugs of rubber-like consistency that obstruct the bronchial tree.[1] Although it has been mostly described in pediatric patients, adult cases have been described following cardiopulmonary bypass, heart surgery, lung transplantation, and sickle cell crisis. Plastic bronchitis has also been described in association with pulmonary diseases, including severe asthma, aspergillosis, bronchiectasis, and cystic fibrosis.[2] Because of different pathophysiologies, two distinct types of casts have been pathologically described, where type 1 casts are richly cellular and associated with inflammatory conditions and type 2 casts are mostly mucinous and scarcely cellular.[3]

Pathogenesis of bronchial cast formation remains poorly understood and depends on multiple factors, including airway inflammation, abnormal lymphatic drainage of the bronchial tree, both congenital or acquired following cardio-thoracic surgery, and any other acute pathology that can trigger inflammation in the lung. Abnormal lymphatic drainage may be caused by damage to the lymphatic structures from the surgical dissection or from position of the bypass cannulas. To our knowledge, two other cases of plastic bronchitis following cardiac surgery in adults have been reported.[4,5]

Plastic bronchitis presents with signs of respiratory distress related to acute airway obstruction by endobronchial casts. Severity of symptoms depends on severity of airway obstruction, including cough, solid expectoration, wheezing, chest pain, fever, acute respiratory distress leading to poor oxygen saturation, and death. Mortality has been described in up to 60% of cases.[6]

Diagnosis of plastic bronchitis is clinical with confirmation on bronchoscopic examination of the respiratory tract. Pathologic examination of endobronchial casts shows mucinous material devoid of inflammation and scarcely cellular. Casts associated with inflammatory conditions (asthma, cystic fibrosis, sickle cell diseases) are rich in inflammatory cells, fibrin, and crystals.

Ideal management of plastic bronchitis is debated. In severe cases, the challenge is to clean and clear the bronchial tree while providing adequate respiratory support. Flexible or rigid bronchoscopy can be useful to remove endobronchial casts, but endoscopic procedures can be long and necessitate adequate pulmonary reserve. Use of mucolytic agents such as n-acetylcysteine, hypertonic saline, or direct endotracheal instillation of

tissue plasminogen activator or recombinant human DNase have been described without proven benefit.[1] Use of ECMO support can be lifesaving in patients presenting with severe respiratory failure and can allow bronchial cleaning until adequate lung function can be restored.

We describe a rare case of plastic bronchitis following a Ross procedure. Whether the precipitating factor leading to acute bronchial obstruction of the airways was related to an immune reaction triggered by the cardiac surgery, abnormal lymphatic drainage related to position of the cardiopulmonary bypass cannulas, blood exposure to the cardiopulmonary bypass circuit, or the human cadaveric valve used for pulmonary reconstruction remains uncertain. Use of ECMO support in this patient was lifesaving and allowed complete airway cleaning with repeated flexible bronchoscopy and use of mucolytic agents.

Acknowledgements The authors sincerely thank Drs Martin Chevrier and Maxime St-Amant for their help with clinical images.

Author contribution *Mohamed Abdel Halim*, and *Michael Mayette* contributed to the writing and approval of the manuscript.

Disclosures None.

Funding statement None.

Editorial responsibility This submission was handled by Dr. Philip M. Jones, Deputy Editor-in-Chief, *Canadian Journal of Anesthesia/ Journal canadien d'anesthésie.*

References

1. *Ntiamoah P, Mukhopadhyay S, Ghosh S, Mehta AC.* Recycling plastic: diagnosis and management of plastic bronchitis among adults. Eur Respir Rev 2021; DOI: https://doi.org/10.1183/16000617.0096-2021.
2. *Feray S, Mora P, Decavele M, Pham T, Hafiani EM, Fartoukh M.* Plastic bronchitis: an unusual complication of acute chest syndrome in adult. Respir Med Case Rep 2017; 21: 93-5.
3. *Seear M, Hui H, Magee F, Bohn D, Cutz E.* Bronchial casts in children: a proposed classification based on nine cases and a review of the literature. Am J Respir Crit Care Med 1997; 155: 364-70.
4. *Sheikh AY, Ahmadi-Kashani M, Mohindra V, Friedenberg A, Pramanik SB, Ogden WD.* A rare case of plastic bronchitis in an adult patient after cardiopulmonary bypass. Ann Thorac Surg 2016; 101: 1176-8.
5. *Schmitz L, Rihawi M.*Plastic bronchitis: a complication of myocardial revascularization. Am J Respir Crit Care Med 2012; 185: 896-7.
6. *Coen M, Daniel L, Serratrice J.* An adult case of plastic bronchitis: a rare and multifactorial disease. J Thorac Dis 2018; 10: E16-9.

Publisher's Note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.