



Multidisciplinary Team Repair of a Primary Aorto-bronchopulmonary Fistula in a Patient with Chronic Type B Aortic Dissection

Shruthi Nammalwar, MD^{1*}, Aamir Shah, MD^{2,5}, Daniel Delgadillo, MD¹, Cassra Arbabi, MD^{1,2}, Edward Ray, MD³, Taryne Imai, MD⁴, Ali Azizzadeh, MD^{1,2}

¹Department of Surgery, Cedars-Sinai Medical Center, Los Angeles, CA

²Division of Vascular Surgery, Cedars-Sinai Medical Center, Los Angeles, CA

³Division of Plastic and Reconstructive Surgery, Cedars-Sinai Medical Center, Los Angeles, CA

⁴Division of Thoracic Surgery, Cedars-Sinai Medical Center, Los Angeles, CA

⁵Department of Cardiac Surgery, Cedars-Sinai Medical Center, Los Angeles, CA

***Corresponding author:** Shruthi Nammalwar, MD, Resident, Department of Surgery, Cedars-Sinai Medical Center 127 S. San Vicente Blvd, Suite A3100, Los Angeles, CA 90048. Tel: (424) 315- 4551; Email: Shruthi.Nammalwar@cshs.org

Citation: Nammalwar S, Shah A, Delgadillo D, Arbabi C, Ray E, et al. (2021) Multidisciplinary Team Repair of a Primary Aorto-bronchopulmonary Fistula in a Patient with Chronic Type B Aortic Dissection. Ana Surg Surgi Cas Rep: ASSCR: 116.

Received Date: 10 December, 2021; **Accepted Date:** 20 December, 2021; **Published Date:** 27 December, 2021

Abstract

Aorto-bronchopulmonary fistula (ABPF) is an exceedingly rare phenomenon and often a postmortem diagnosis. Early diagnosis and expedited surgical intervention contribute to successful outcomes. Here we describe a multidisciplinary approach to the open repair of a primary ABPF.

Keywords: Aorto-bronchopulmonary fistula, aortic dissection, cardiopulmonary bypass, thoracoabdominal, multidisciplinary.

Introduction

Aorto-bronchopulmonary fistula (ABPF) is a rare phenomenon which invariably tends to be lethal if it is not diagnosed and intervened upon expeditiously.¹ ABPF is defined as a connection between the aorta and the bronchopulmonary tree, which was first described in the literature by Keefer et al in 1934.² The etiological factors of ABPF's include aortic dissection, aneurysm, infection, lung malignancy, lung transplantation, trauma, and surgical interventions with or without prosthesis.³ It has been recommended that the clinical presentation of massive hemoptysis should warrant a high index of suspicion of ABPF and further investigation.^{4,5} Although, most cases of ABPF's are encountered at autopsy⁴, here we describe a multidisciplinary approach and successful open repair of a primary ABPF.

Case Report

A 65-year-old man who had a history of uncontrolled hypertension and prior acute type A aortic dissection for which he had undergone ascending aorta and hemiarch replacement at an outside facility presented with a chronic residual type B dissection and massive hemoptysis. He had Computerized Tomography Angiography (CTA) of the chest which revealed a new periaortic hematoma with adjacent inflammatory changes in the lung parenchyma that was concerning for aortopulmonary fistula (Figure 1). A multidisciplinary reconstructive approach was taken with the assistance of various surgeons including a plastic surgeon, a thoracic surgeon, a vascular surgeon and a cardiac surgeon.

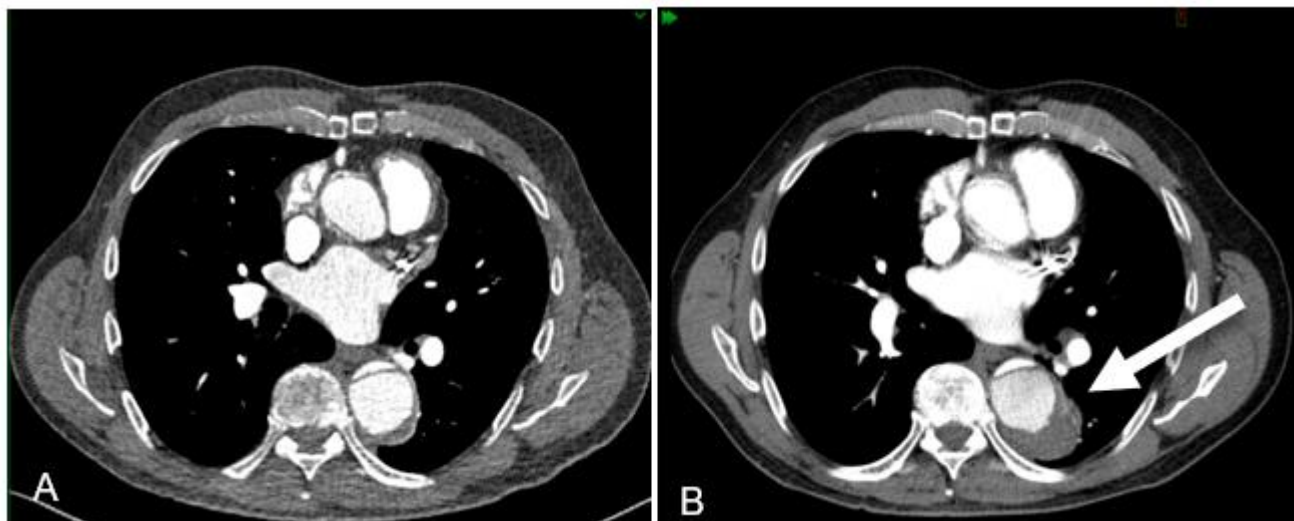


Figure 1: CTA of the chest from two months prior to admission shows the patient's chronic type B dissection (A). CTA of the chest on admission reveals the new ABPF with a periaortic hematoma and adjacent inflammatory changes (B).

The patient was taken as an emergency to the operating room and he underwent general endotracheal anesthesia. Flexible bronchoscopy was undertaken which revealed old blood within his trachea without active bleeding from the left lung. Surgical repair was undertaken through a thoracoabdominal approach via the left 5th intercostal space. A latissimus dorsi pedicled flap was prepared by the plastic surgeon prior to entry into the thoracic cavity. Cardiopulmonary bypass (CPB) via left femoral artery and vein cannulation was initiated with systemic hypothermia to

20° centigrade. Extensive adhesions were encountered between the visceral pleura, mediastinum and chest wall. The lower lobe of the left lung was densely adherent to the mid-thoracic aorta near the ABPF. Circulatory arrest was initiated, and the lower thoracic aorta was clamped to enable distal aortic perfusion. The ABPF was dissected free, and a 2 cm connection between the left lower lobe and false lumen of the chronic aortic dissection was confirmed (Figure 2).

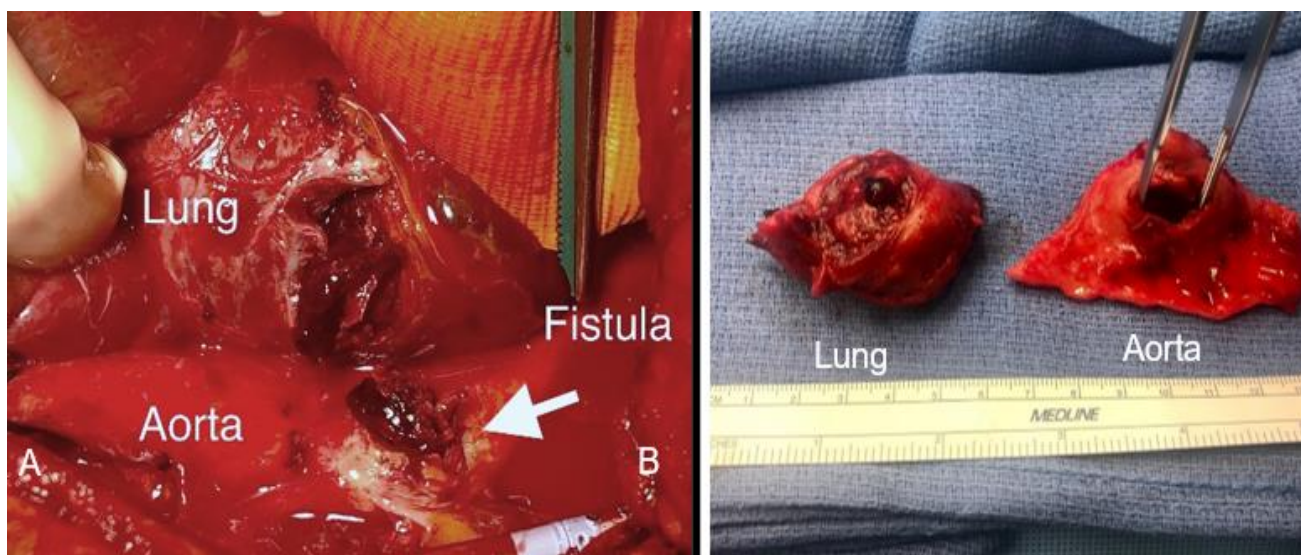


Figure 2: Careful dissection delineates the 2 cm fistula between the left lower lobe and the false lumen of the chronic aortic dissection (A). The aorta was transected distal to the left subclavian artery origin and distal to the fistula while a left lower lobe wedge containing the fistula was resected. Resection of the ABPF fistula was confirmed (B).

The aorta was transected distal to the origin of the left subclavian artery and proximal fenestration of the septum was performed into the aortic arch. A 32mm Rifampin soaked Gelweave™ Ante-Flo™ (Terumo, Tokyo, Japan) graft with a reversed elephant trunk was utilized for the proximal anastomosis. Central arterial cannulation was then carried out via the side branch of the aortic graft, and flow was restored to the upper body. Systemic rewarming was

initiated, and the distal anastomosis was completed. A wedge resection of left lower lobe pulmonary fistula was performed by a thoracic surgeon. The latissimus dorsi flap was set into the chest cavity via the third intercostal space and wrapped around the aortic graft for protection (Figure 3). The patient was weaned from CPB and the wound was closed after placing thoracostomy tubes.

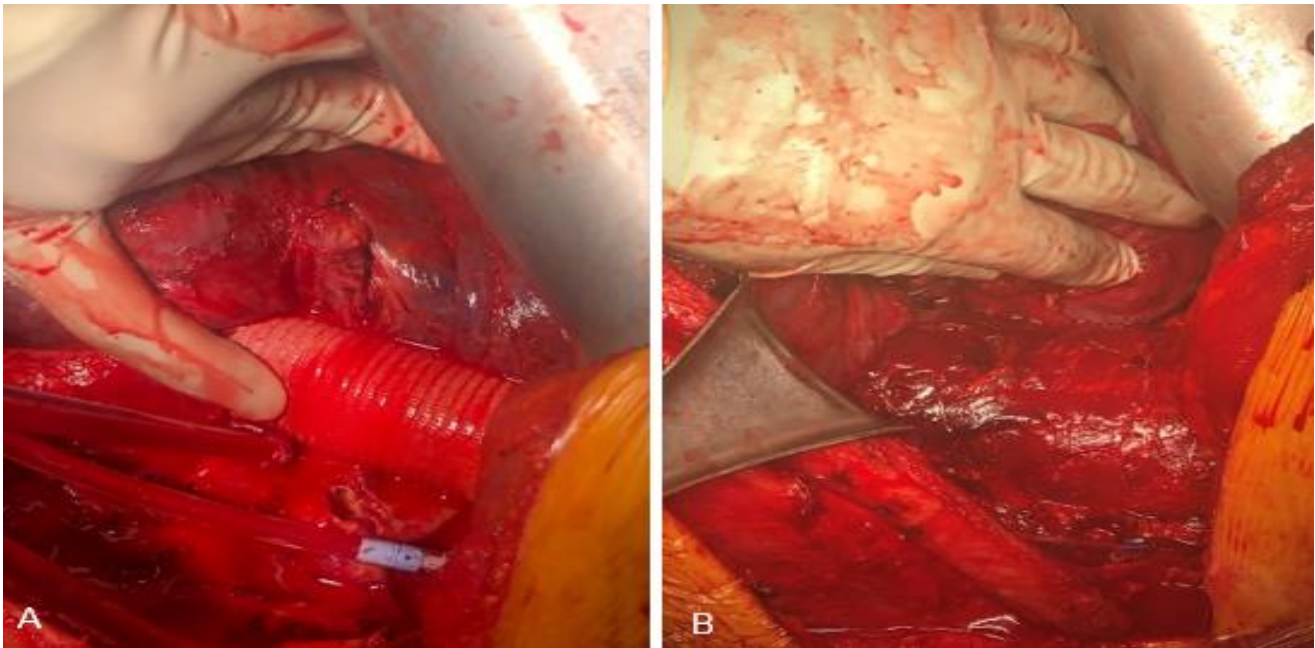


Figure 3: A 32 mm Rifampin soaked Gelwave TM Ante-Flo TM (Terumo, Tokyo, Jpan) graft with a reversed elephant trunk was utilized (A). The latissimus dorsi flap is wrapped over the aortic graft (B).

The patient's convalescence was without significant complications. Mechanical ventilation was discontinued on the first post-operative day. Bacterial cultures of the lung yielded no growth, and the patient was discharged on the 14th post-operative day in stable condition. He was maintained on antibiotic therapy for 6 weeks and continued to progress well at 6 month and 1 year follow-ups with no complications.

Discussion

It has been iterated that the incidence of thoracic aortic dissection is likely underestimated at 3–4 cases per 100,000 persons per year, with the incidence of primary ABPF substantially less.⁶ Two common theories to explain the pathophysiology of ABPFs are local infection and or mechanical stress leading to fistula formation.⁷ Clinical signs of intermittent hemoptysis may be present and these have been explained by the thrombogenicity of the lung parenchyma. However, it has been stated that massive hemoptysis which is described as greater than 400mL of expectorant in 24 hours is often the expiring event.⁸ Having a high index of suspicion for ABPF is critical as the diagnosis can be challenging given the rare incidence of ABPF and broad differential diagnoses for hemoptysis. Thorough review of the past medical and surgical history along with high quality radiology imaging is paramount for the establishment of this diagnosis. The use of radiology imaging such as CTA along with the technological advancements has greatly contributed to the diagnosis.⁹ Once an ABPF is diagnosed, it is crucial to establish a multidisciplinary team that is prepared to manage this complex and often fatal condition.

Repair of a primary ABPF is at risk of aortic prosthetic graft infection. Typically, conservative management with antibiotics or surgical debridement are ineffective in the long-term. A pedicled flap can be a valuable prophylactic

tool to minimize postoperative infections and to improve patient outcomes.^{12,13} Latissimus dorsi flaps are routinely used in thoracic surgery for bronchopleural fistula repairs and esophageal repair.¹⁴ An intrathoracic muscle transposition can be valuable to buttress aortic prosthetic grafts to avoid local wound infection.¹² In addition, other pedicled flaps, including pectoralis major, rectus abdominis, and serratus anterior flaps, can be used to reinforce the aortic repair.

Strong considerations should be placed on the patient's clinical stability, risk factors, available medical expertise, and treatment modalities. In an unstable or high operative risk patient, endovascular treatment with an aortic stent-graft is a fast and minimally invasive approach that can be a temporizing measure or can be a definitive treatment; however, with an incurred risk of >10% for adverse sequelae.^{10,11} Thus, in a stable patient with appropriate risk, open surgery as described above utilizing a multidisciplinary approach could provide for a more optimal definitive management.

Conclusion

Primary ABPF is a rare phenomenon, and it is uniformly fatal if surgically untreated. A high index of suspicion and early recognition are essential and can lead to improved outcomes. Any patient with aortic pathology and hemoptysis should be evaluated for aortopulmonary fistula. A multidisciplinary approach can result in a life-saving procedure in the face of a challenging presentation.

Conflict of interest

No conflicts of interest to declare.

Acknowledgements

The successful repair of this ABPF would not have been possible without the multidisciplinary team. We would like to thank the intensive care unit staff, the intensivists, the

radiologists, the department of thoracic surgery, the department of plastic surgery, the department of vascular surgery, the department of cardiac surgery team, and the operating room staff for contributing to this patient's successful care.

References

1. Li M, Langlois N, Byard RW. Fatal aortobronchial fistula. *J Forensic Leg Med*. 2013;20(5):395-398.
2. Keefer C, Mallory K. The pulmonary and pleural complications of aortic aneurysm. *American Heart Journal*. 1934;10(2):208-220.
3. Maffei V, Simmini S, Rossato M, Fioretto P, Fallo F, Basso C, Rizzo S. Sudden death with massive hemoptysis from aortobronchial fistula. *Cardiovasc Pathol*. 2020; 44:107158.
4. Coblenz CL, Sallee DS, Chiles C. Aortobronchopulmonary fistula complicating aortic aneurysm: diagnosis in four cases. *AJR Am J Roentgenol*. 1988;150(3):535-538.
5. Fernández González AL, Montero JA, Luna D, Gil O, Sanjuán VM, Monzonís AM. Aortobronchial fistula secondary to chronic post-traumatic thoracic aneurysm. *Tex Heart Inst J*. 1996;23(2):174-177.
6. LeMaire SA, Russell L. Epidemiology of thoracic aortic dissection. *Nat Rev Cardiol*. 2011;8(2):103-113.
7. MacIntosh EL, Parrott JC, Unruh HW. Fistulas between the aorta and tracheobronchial tree. *Ann Thorac Surg*. 1991;51(3):515-519.
8. Demeter SL, Cordasco EM. Aortobronchial Fistula: Keys to Successful Management. *Angiology*. 1980;31(7):481-487.
9. Hampson S, Pepper J. Aortopulmonary fistula: role of computed tomography. *Thorax*. 1987;42(5):395-396.
10. Kokotsakis J, Misthos P, Athanasiou T, Romana C, Skouteli E, Lioulis A, Kaskarelis I. Endovascular stenting for primary aortobronchial fistula in association with massive hemoptysis. *Tex Heart Inst J*. 2007;34(3):369-372.
11. Nguyen T, Peters P, McGahan T, Shah P. Staged management of a primary aortobronchial fistula: a novel approach using a trapezius flap repair. *Heart Lung Circ*. 2012;21(5):292-294.
12. Taguchi S, Mori A, Suzuki R, Ishida O. Technique for using pedicled latissimus dorsi muscle flaps to wrap prosthetic grafts in an infected thoracic aorta. *Ann Vasc Surg*. 2013 Nov;27(8):1223-1227.
13. Frautschi RS, Bassiri Gharb B, Duong MM, Gurunluoglu R, Papay F, Zins JE, Rampazzo A. The Cardioplastic Approach to the Treatment of Infected Aortic Grafts. *Ann Plast Surg*. 2017 Aug;79(2):221-225.
14. Abolhoda A, Bui TD, Milliken JC, Wirth GA. Pedicled latissimus dorsi muscle flap: routine use in high-risk thoracic surgery. *Tex Heart Inst J*. 2009;36(4):298-302.