

Massive hemoptysis secondary to Behçet's aneurysm controlled with endobronchial balloon lung isolation: Case report and review of literature

Ali Al Bshabshe^{a,*}, Amer Assiri^b, Mansour Somaily^c

^a Department of Internal Medicine, Critical Care Division, College of Medicine, King Khalid University, Abha, Saudi Arabia

^b Department of Medicine Division of Critical Care, King Khalid University Medical City, Abha, Saudi Arabia

^c Department of Medicine Rheumatology Division, King Khalid University Medical City, Abha, Saudi Arabia

ARTICLE INFO

Keywords:

Behçet's disease
Massive hemoptysis
Fiberoptic bronchoscopy
Endobronchial balloon
Lung isolation

ABSTRACT

Behçet's disease (BD) is a multisystem disorder with various clinical presentations. Herein, we present a lethal complication associated with pulmonary artery aneurysm due to BD, resulting in massive hemoptysis that was controlled using endobronchial balloon lung isolation.

1. Introduction

Behçet's disease (BD) is a multisystem disorder, first described by Hulusi Behçet in 1937. It is a vasculitis that presents with a triad of recurrent ulcers of the oral and genital mucosa with relapsing uveitis. Among the systemic vasculitides, BD is remarkable for its ability to involve blood vessels of all sizes (small, medium, and large) on both the arterial and venous sides of the circulation [1]. The most commonly affected arteries are the aorta, pulmonary, and femoral arteries [2].

2. Case report

The patient was a 45 year old man, with a history of Behçet's disease for six years that was complicated by recurrent episodes of deep vein thrombosis, pulmonary embolism (Fig. 1), and a left pulmonary artery aneurysm (PAA) for which he had undergone coil embolization two months prior to presentation to our hospital. (Figs. 2 and 3). He had multiple episodes of mild hemoptysis over three days that was associated with shortness of breath and chest pain for which he did not seek medical attention. He then had one episode of massive hemoptysis (around 1200 ml of fresh blood), and was brought to our hospital. He became agitated and hypoxic (Spo₂, 75% on 15 L non-rebreathing face mask) and required immediate intubation and mechanical ventilation.

He continued to have hemoptysis and hypoxemia (Spo₂, 60%) on 100% FiO₂. An emergency fiberoptic bronchoscopy was undertaken that showed fresh blood coming from the left main stem bronchus along with

multiple clots. The left lung was isolated using endobronchial balloon occlusion (Fig. 4). Blood clots were removed from the right bronchial tree using forceps after which his saturation improved, and he was admitted to the critical care unit. The endobronchial balloon was deflated 24 hours later and the airway was assessed by fiberoptic bronchoscopy to ensure no further bleeding. A definitive surgical intervention was planned for the patient by the consulting cardiothoracic team. He was subsequently extubated and discharged from the intensive care unit in a stable condition.

3. Discussion

BD is a rare multisystem disorder of unknown etiology presenting with recurrent oral and/or genital ulcerations and chronic relapsing uveitis that may cause blindness and/or neurological impairments [3–5]. The vascular system is involved in 20–40% of patients with BD, with the venous circulation more commonly involved than the arterial system [2].

PAA is the most common lung manifestation of BD, and interestingly, BD is the most common cause of PAAs. Parenchymal findings including atelectasis, hemorrhage, and infarction (focal or sub-pleural consolidation) are commonly encountered secondary to thrombosis of the pulmonary vessels. Additionally, pneumonia, bronchitis, fibrosis, sub-pleural infiltrates, sub-pleural nodules, pleural effusion, and emphysema have been also been described [6].

Massive hemoptysis has multiple definitions, but the most plausible

* Corresponding author.

E-mail addresses: albshabshe@yahoo.com (A. Al Bshabshe), amerasiri80@gmail.com (A. Assiri), dr_somaily@yahoo.com (M. Somaily).

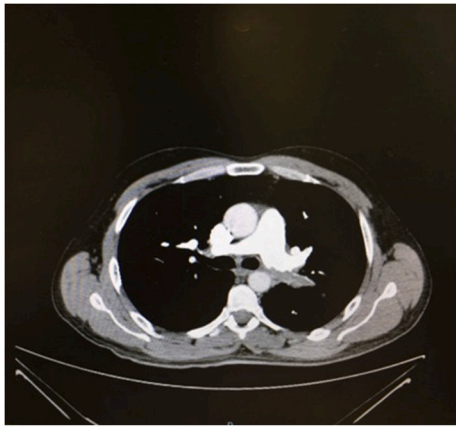


Fig. 1. CT CHEST showing chronic left pulmonary artery thrombus.

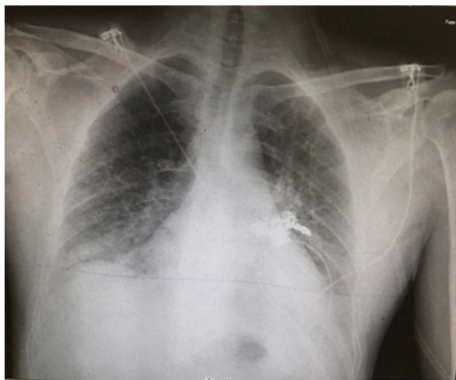


Fig. 2. CXR coils in the left pulmonary artery aneurysm.



Fig. 3. CT CHEST, coils in the left pulmonary artery aneurysm.

one is 'the volume that is life-threatening, either leading to airway obstruction or blood loss.' [7] The hemoptysis seen in BD is usually associated with aneurysms of the pulmonary artery [8]. The use of endobronchial balloons is effective in the management of massive hemoptysis, as it enables the isolation of a bleeding segment of the airway and prevents the aspiration of blood into the larger airways, thereby maintaining airway patency and oxygenation [9]. The balloon must be kept inflated for 24–48 h for the clot to form, although it can be left in the airway for up to several days. It must be regularly deflated, at least 3

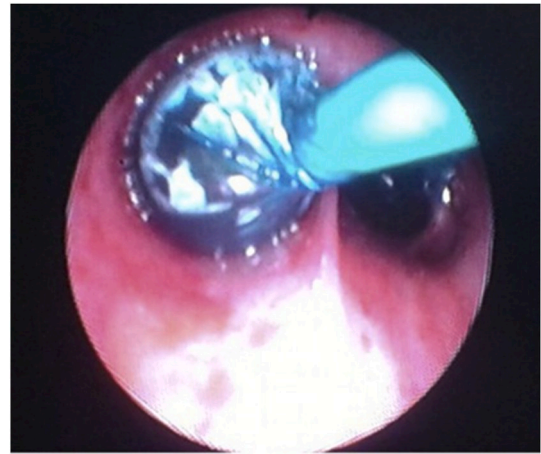


Fig. 4. Bronchoscopy view of left main stem bronchus isolation using endobronchial balloon.

times a day, to prevent ischemia of the mucosa [10].

Generally, the management of hemoptysis is focused on controlling and stopping the bleeding and differentiating massive from non-massive hemoptysis.

Multiple treatment modalities are available and the choice of therapy is based on each clinical scenario to control the hemoptysis. These modalities include, but are not limited to: (1) rigid or fiberoptic bronchoscopy that can facilitate multiple therapeutic options such as laser or argon plasma coagulation, endobronchial stent tamponade, Endobronchial airway blockade, electrocautery, cryotherapy, and brachytherapy; (2) bronchial artery embolization; and (3) surgical intervention [11–15]. Surgical lobectomy is an option when a definitive solution is required for massive hemoptysis due to pulmonary artery aneurysms.

The urgent clinical situation of this patient required a quick and effective solution, and neither rigid bronchoscopy, angioembolization, nor a surgical intervention were viable options at that point. Single lung isolation and clot removal using fiberoptic bronchoscopy enabled us to stabilize the patient and gain time until definitive surgery could be planned.

4. Conclusion

Massive hemoptysis is a life-threatening respiratory emergency and physicians and intensivists need to have multiple options to control it. Bronchoscopic endobronchial balloon isolation is a treatment modality that is feasible, beneficial and can be life-saving.

Statement of informed consent

Informed consent was obtained from the patient for the publication of this report and the accompanying images.

Author contributions

All authors have equally contributed to the conception and manuscript preparation of this case report. All authors have read and approved the final manuscript.

Declaration of competing interest

The authors declare that they have no competing interests.

Acknowledgements

none.

References

- [1] F. Erkan, A. Gul, E. Tasali, Pulmonary manifestations of Behçet's disease, *Thorax* 56 (2001) 572–578, <https://doi.org/10.1136/thorax.56.7.572>.
- [2] I. Guler, K. Odev, R. Tunc, Partially thrombosed multiple pulmonary artery aneurysms secondary to Behçet disease, *Canc. Res. J.* 22 (2) (2015) 75–76, <https://doi.org/10.1155/2015/946425>.
- [3] N. Hiller, S. Lieberman, T. Chajek-Shaul, J. Bar-Ziv, D. Shaham, Thoracic manifestations of Behçet disease at CT, *Radiographics* 24 (2004) 801–808, <https://doi.org/10.1148/rg.243035091>.
- [4] F. Erkan, Pulmonary involvement in Behçet disease, *Curr. Opin. Pulm. Med.* 5 (1999) 314–318, <https://doi.org/10.1097/00063198-199909000-00009>.
- [5] A. Kaya, C. Ertan, O.U. Gurkan, S. Fitoz, C. Atasoy, M. Kilickap, et al., Behçet's disease with right ventricle thrombus and bilateral pulmonary artery aneurysms—a case report, *Angiology* 55 (2004) 573–575, <https://doi.org/10.1177/000331970405500516>.
- [6] N. Ceylan, S. Bayraktaroglu, S.M. Erturk, R. Savas, H. Alper, Pulmonary and vascular manifestations of Behçet disease: imaging findings, *AJR Am. J. Roentgenol.* 194 (2010) 158–164, <https://doi.org/10.2214/AJR.09.2763>.
- [7] A.A. Garzon, M.M. Cerruti, M.E. Golding, Exsanguinating hemoptysis, *J. Thorac. Cardiovasc. Surg.* 84 (1982) 829–833.
- [8] Salamon, A. Weinberger, M. Nili, I. Avidor, M. Neuman, A. Zelikovsky, M.J. Levy, J. Pinkhas, Massive hemoptysis complicating behçet's syndrome: the importance of early pulmonary angiography and operation, *Ann. Thorac. Surg.* 45 (1988) 566–567, [https://doi.org/10.1016/S0003-4975\(10\)64538-9](https://doi.org/10.1016/S0003-4975(10)64538-9).
- [9] J.L. Lordan, A. Gascoigne, P.A. Corris, The pulmonary physician in critical care c Illustrative case 7: assessment and management of massive hemoptysis, *Thorax* 58 (2003) 814–819, <https://doi.org/10.1136/thorax.58.9.814>.
- [10] L. Freitag, E. Tekolf, G. Stamatis, et al., Three years experience with a new balloon catheter for the management of hemoptysis, *Eur. Respir. J.* 7 (1994) 2033–2037.
- [11] J.Y. Chun, R. Morgan, A.M. Belli, Radiological management of hemoptysis: a comprehensive review of diagnostic imaging and bronchial arterial embolization, *Cardiovasc. Intervent. Radiol.* 33 (2010) 240–250, <https://doi.org/10.1007/s00270-009-9788-z>.
- [12] W.M. Edmondstone, E.M. Nanson, A.A. Woodcock, F.J. Millard, M.R. Hetzel, Life threatening haemoptysis controlled by laser photocoagulation, *Thorax* 38 (1983) 788–789, <https://doi.org/10.1136/thx.38.10.788>.
- [13] L. Sakr, H. Dutau, Massive hemoptysis: an update on the role of bronchoscopy in diagnosis and management, *Respiration* 80 (2010) 38–58, <https://doi.org/10.1159/000274492>.
- [14] H. Ittrich, M. Bockhorn, H. Klose, M. Simon, The diagnosis and treatment of hemoptysis, *Dtsch Arztebl Int.* 114 (21) (2017) 371–381, <https://doi.org/10.3238/arztebl.2017.0371>.
- [15] R. Cordovilla, E. Bollo de Miguel, A. Nun ãez Ares, F.J. Cosano Povedano, I. Herráez Ortega, R. Jiménez Merchán, Diagnosis and treatment of hemoptysis, *Arch. Bronconeumol.* 52 (2016) 368–377, <https://doi.org/10.1016/j.arbres.2015.12.002>.