

Case Report

Mediastinal bronchial artery aneurysm treated with aortic stent and embolization: case report

Meijun Song, Hongcheng Wu, Jingbo Jiang, Guo'an Wang, Shibo Wu, Ting Ge

Department of Respiratory Medicine, Ningbo Medical University Affiliated Li Huli Hospital, Ningbo, China

Received February 24, 2016; Accepted August 15, 2016; Epub September 15, 2016; Published September 30, 2016

Abstract: Mediastinal bronchial artery aneurysm (BAA) refers to a rare aneurysmal dilatation but can be potentially life threatening. Opportune treatment is mandatory upon a confirmed diagnosis. There are several reports of endovascular treatment of BAA with transcatheter arterial embolization (TAE) and only a few cases treated with aortic stent-graft exclusion. Here, we report on two case studies of mediastinal BAAs with a short neck treated with a combined approach of stent-graft occlusion of the inflow and coil embolization of the outflow arteries. As a comparison, we reviewed seven other cases that were previously reported in literature for their clinical presentation and therapeutic management. Our study demonstrated that mediastinal BAA can be successfully treated with the available endovascular techniques. Combining approaches of aortic stent-graft placement and coil embolization is a viable option in the treatment of BAA with a short neck.

Keywords: Aneurysm, angiography, embolization, covered stent-graft, thoracic aorta

Introduction

The occurrence of bronchial artery aneurysm (BAA) is rare and detected in less than 1% of all patients who undergo selective bronchial arteriography [1]. BAA can be intrapulmonary, mediastinal or both, and the clinical presentation depends on the size, location, and presence of concomitant disease. The main indications of mediastinal BAA is related to compression or rupture into contiguous structures [2]. Its rupture can cause a life-threatening hemorrhage and opportune treatment is mandatory upon a confirmed diagnosis. Open surgical procedures are preferred but recently transcatheter arterial embolization (TAE) has demonstrated good treatment results [3]. Currently, there are numerous reports on endovascular treatment of BAA using TAE and a few cases were treated with aortic stent-graft exclusion [3-5]. Here, we report on 2 cases of mediastinal BAAs treated with a combined approach of stent-graft occlusion of the inflow and coil embolization of the outflow arteries. As a comparison, we also reviewed seven other cases previously reported in literature and compared them with our current case studies.

Case report

CASE 1

A 59-year-old woman was admitted to the Emergency Room due to a sudden onset of chest pain radiating to back, with a history of constant dysphagia lasting for about 3 years. She also had a history of repeated hemoptysis for more than 10 years. Upon reaching the hospital, the patient was in hemodynamically stable condition except for mild elevation of her respiration rate (22/min); her blood pressure was 118/76 mmHg, pulse rate was 67 beats/min, and the heart rhythm was regular. An EKG presented a normal sinus rhythm without any ischemic signs or arrhythmia. Breath sounds were normal. Laboratory examination revealed hemoglobin of 12.5 g/dl. There was no trauma, and no sign of infection.

A chest computed tomographic (CT) scan (**Figure 1**) confirmed posterior mediastinum hematoma and bilateral bronchiectasis. Because of a suspicious origin of mediastinal hemorrhage on the chest CT scan, thoracic aortogram and bronchial arteriogram were con-

Treating BAA with aortic stent and embolization

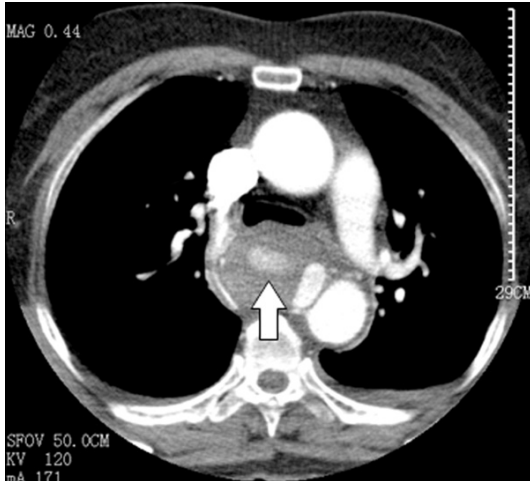


Figure 1. Enhanced CT scan showed posterior mediastinum hematoma.

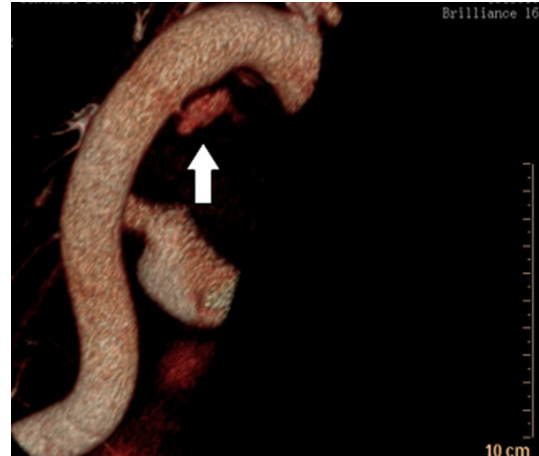


Figure 3. CT scan and 3D reconstruction confirmed a bronchial artery aneurysm (18 mm) in the initial part of the bronchial artery, with a neck of 5 mm in length.

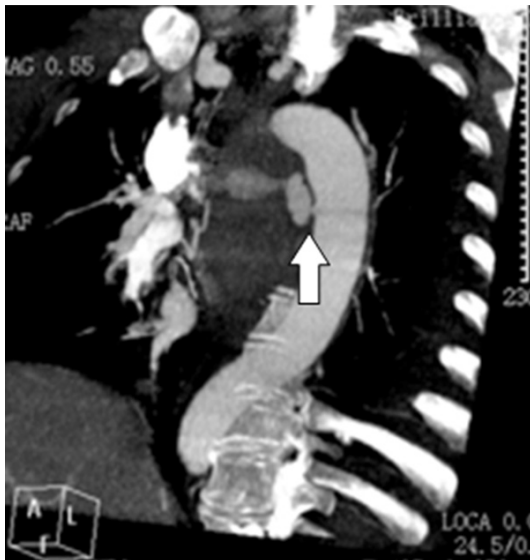


Figure 2. CT scan and 3D reconstruction confirmed a bronchial artery aneurysm (18 mm) in the initial part of the bronchial artery, adjacent to the descending aorta.

ducted. On the selective bronchial arteriogram, we found a bronchial artery aneurysm (18 mm) that was just in the initial part of the bronchial artery, located very close to the aorta as shown in **Figures 2** and **3**. The length of the neck was about 5 mm. There was no sign of active bleeding or extravasation. To prevent further rupture or extravasation of the aneurysm, we performed a bronchial artery embolization with several microcoils. After that, the second time of selective bronchial arteriogram showed the dis-



Figure 4. After bronchial artery embolization with several microcoils, selective bronchial arteriogram showed the disappearance of the distal end of the bronchial artery but the initial part presented aneurysmal dilatation.

appearance of the distal end of the bronchial artery but we still found the initial part presented aneurysmal dilatation (**Figure 4**). Then an aortic stent was planted in the descending aorta to isolate the bleeding bronchial artery, and bronchial artery angiography showed the blood flow was completely blocked (**Figure 5**).

The patient was in a stable condition during percutaneous vascular intervention. She was sent to the ICU and under close observation after the embolization procedure. During follow-up on the first month after operation, CT scan of the chest showed left pulmonary atel-

Treating BAA with aortic stent and embolization



Figure 5. An aorta stent was planted in the descending aorta, bronchial artery angiography showed disappearance of the BAA but microcoils.



Figure 6. In follow-up of the third month after operation, CT scan of the chest showed no stent migration, and disappearance of the BAA.

ectasis and pleural infusion. A chest tube was inserted and 2.5 liters of unclotted blood was drained from the left pleural cavity. After that, a subsequent CT scan showed pulmonary re-expansion and no further bleeding, indicating her conditions returned to normality. In the



Figure 7. Contrast-enhanced computed tomography (CT) scan shows posterior mediastinum hematoma and bilateral pleural effusion, full of blood in the left cavity.



Figure 8. CT scan and 3D reconstruction shows the short neck (6 mm) of the Bronchial artery aneurysm and its origin from the descending aorta.

third month after operation, CT scan of the chest showed no stent migration or pleural effusion as shown in **Figure 6**.

CASE 2

A 63-year-old man was admitted to the Emergency Department due to a sudden onset of sharp chest pain. The pain was located in the anterior chest wall radiating to the back. Upon reaching the hospital, the patient was in hemodynamically unstable condition, his respiration rate was 23/min; blood pressure was 70/40

Treating BAA with aortic stent and embolization

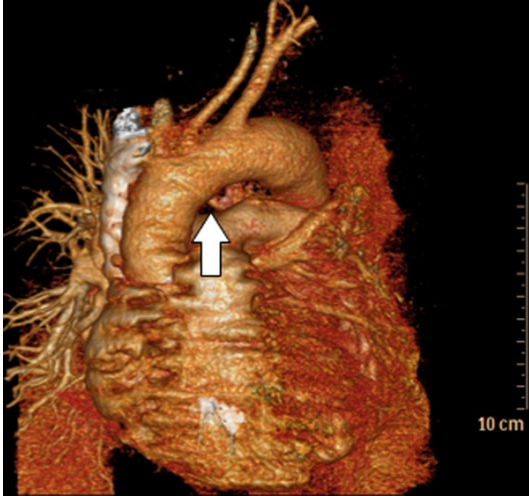


Figure 9. CT scan and 3D reconstruction shows the short neck of the Bronchial artery aneurysm and its origin from the descending aorta.

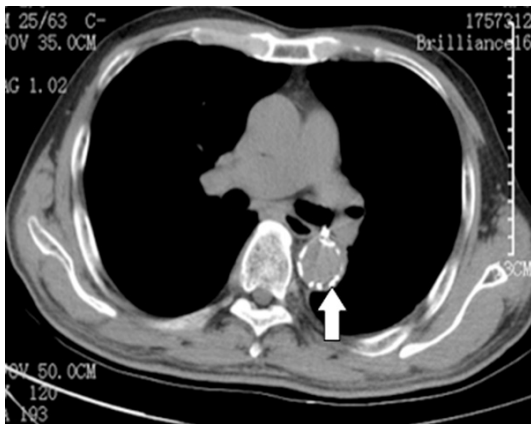


Figure 10. CT scan showed little pleural effusion in the left chest, no shift of the stent 3 month after operation.

mmHg, pulse rate was 200 beats/min, but the heart rhythm was regular. Left breath sounds were very low. Laboratory examination revealed hemoglobin of 8.9 g/dl. There was no trauma, no sign of infection, and no hemoptysis.

A chest CT scan (**Figure 7**) confirmed active posterior mediastinum hematoma that was very close to the descending aorta and bilateral pleural effusion. The left chest cavity was full of blood. A chest tube was inserted into the left pleural cavity. Due to a suspicious origin of mediastinal hemorrhage on the chest CT scan, thoracic aortogram and bronchial arteriogram were conducted. On the selective bronchial ar-

teriogram, we found a bronchial artery aneurysm of 5 mm. The location was situated in the initial part of the bronchial artery and adjacent to the descending aorta, with a 6 mm neck length as shown in **Figures 8** and **9**. There was extravasation of contrast agent in the mediastinum, which strongly suggested the presence of active bleeding. Therefore, we performed a bronchial artery embolization with 3 microcoils. After that, the second time of selective bronchial arteriogram showed the disappearance of the distal end of the bronchial artery. However, we still found some contrast agent in the initial part of the bronchial artery near the descending aorta. Then a stent (140 mm×34 mm) was planted in the descending aorta to isolate the bleeding bronchial artery, and bronchial artery angiography showed the blood flow was completely blocked.

The patient was in a stable condition during percutaneous vascular intervention. He was sent to the ICU for close observation after the embolization procedure. Due to his loss of blood, the patient needed transfusion. During follow-up on the third month after operation, the patient was in good condition without further complications. His chest CT showed no pleural effusion in the left chest and no shift of the stent (**Figure 10**).

Discussion

Acute hemomediastinum and hemothorax are usually related to chest trauma, rupture of a thoracic aortic aneurysm, or aortic dissection [6]. The causes of spontaneous mediastinal hemorrhage can fall within four distinct categories [7] that include: (1) complication of enlarging mediastinal masses; (2) transient increase in intra-thoracic pressure; (3) sudden sustained hypertension; and (4) altered hemostasis. A ruptured BAA is seldom associated and rarely the causation of mediastinal hemorrhage. Mediastinal BAA is a rare condition, which has been reported only in very few cases in the literature [3, 8].

The exact cause of BAA is unclear [1]. The condition can be congenital, as in the context of pulmonary sequestration [9] or pulmonary agenesis [10]. It can also be acquired, due largely to atherosclerosis [11, 12], inflammatory lung disease, bronchiectasis [1], tuberculosis or trauma (pseudoaneurysm) [13]. Other asso-

Treating BAA with aortic stent and embolization

Table 1. Comparison of published cases of ruptured mediastinal bronchial artery aneurysm treated with aortic stent

	Time	Sex/age	Cause	BAA (mm)	Neck description	Region	Symptom	Stent (mm)	Outcome	Hospitalization post operation
Sakai et al. [5]	1998	M/72	Bronchiectasis	25	Near the aorta	Mediastinal	Hemoptysis	NA	Successful	NA
Kasashima, et al. [4]	2003	M/79	Tuberculosis	60	Near the aorta	Mediastinal	Hoarseness	40×200	Successful	15 days
Sanchez E et al. [22]	2007	F/69	Tuberculosis	40	wide	Mediastinal	Dysphagia	30×150	Successful	3 days
Takahashi Y et al. [21]	2010	M/74	Bronchiectasis	60	NA	Mediastinal	Productive Cough	31×150	Successful	15 days
Bouayad M et al. [24]	2011	M/80	Unknown	NA	Adjacent to aorta	Mediastinal	Disphagia	NA	Successful	NA
Hu et al. [23]	2011	M/67	Hypertension	35	NA	Mediastinal, Pleural cavity	Chest pain	NA	Successful	5 days
Guzzardi G et al. [19]	2012	M/66	COPD and cystic bronchiectasis	40	5 mm	Mediastinal	Dyaphagia	30×100	Successful	6 days
Present case 1	2014	F/59	Bronchiectasis	18	5 mm	Mediastinal	Chest pain dysphagia	30×160	Successful	21 days
Present case 2	2014	M/63	Unknown	5	6 mm	Mediastinal, Pleural cavity	Chest pain	34×140	Successful	17 days

M, male; F, female; NA, not available.

Treating BAA with aortic stent and embolization

ciations to BAA include systemic vascular abnormalities, such as Osler-Weber-Rendu disease [14] and sepsis (mycotic aneurysm) [15]. The focal weakening or injury to the vessel walls is the common link among these causes. Increased blood flow to the bronchial arteries may also play a significant role. As shown in **Table 1**, our review of past literature highlighted causes due to bronchiectasis, tuberculosis, cystic bronchiectasis and hypertension. Two of the studies were of unknown etiology. The majority of the causes were linked to chronic inflammation.

Once the bronchial artery ruptures, the clinical presentation is acute and life-threatening, with the most common symptoms being severe chest/back pain mimicking acute aortic dissection [16] together with symptoms of shock [17]. On the other hand, the most common hemorrhagic presentation is hemoptysis, followed by hematemesis, depending on whether the aneurysm extends parenchymally or posteriorly. Massive hemomediastinum is less common and hemothorax is the least common mode of presentation [18]. It may also be accompanied with epigastric pain, hematemesis, and hemoptysis resulting from the rupture into the esophagus and pulmonary parenchyma. Mediastinal BAA may present compressive symptoms, such as dysphagia or superior vena cava syndrome [12]. Although little is known about the process that leads to BAA rupture, the size of the aneurysm (5-40 mm) is not an incremental risk factor [3]. Prior studies have reported on radiological examination of BAAs with sizes as large as 8-10 cm in diameter, which were not as serious as cases of BAAs with sizes of 2 cm. Patients in the latter cases were manifested with life threatening bleeding conditions. As observed also in **Table 1**, all cases with ruptured BAAs were of relatively small size ranging from 5-60 mm in diameter. This supports that the size of the aneurysm does not negatively correlate to clinical outcome.

The primary diagnostic modes are computed tomographic angiography and intra-arterial angiography. Occasionally, magnetic resonance imaging has been employed in complex cases. Here we describe mediastinal BAA with a short neck. We compared our study against other reported cases (**Table 1**) but most studies lack comprehensive quantitative data of the neck

description. Comparing with the study conducted by Guzzardi G *et al.* [19], our results were consistent with their findings of measured neck length. Our measured neck length for case 1 and 2 were 5 mm and 6 mm respectively.

BAA should be treated whether it is symptomatic or not. In previous reports, TAE has been the most common approach in recent years [3]. TAE to occlude the afferent and efferent arteries of the BAA is considered the first line treatment option if the patient is stable. Surgery should be reserved for patients with contraindications to embolization, such as allergy to iodinated contrast medium or medullary artery. Open surgical treatment is a valid alternative but is associated with high morbidity and mortality. The advantages and disadvantages of surgery and TAE should be recognized, and the appropriate procedure should be selected based on the patient's clinical status. Several reports commented on the failed embolization of a bronchial artery if the origin of the aneurysm is too close to the aorta [7, 20]. Given the clinical presentation, the optimal treatment for mediastinal BAA especially for that with short neck is combined treatment with TAE and aortic stent. Kasashima *et al.* [4] reported successful treatment of BAA by stent-graft placement alone. Sakai *et al.* [5] Takahashi *et al.* [21] and Giuseppe Guzzardi *et al.* [19] reported three cases with the treatment of BAA by stent-graft occlusion of the inflow artery and coil embolization of the outflow artery, demonstrating the effectiveness of this kind of combined approach. If transcatheter coil embolization of the outflow vessels is technically difficult, it is possible to embolize these arteries using fibrin sealant, as described by Sanchez *et al.* [22]. Hu *et al.* [23] described successful combined treatment of BAA with aortic stent-graft placement and embolization of the outflow arteries using sodium polymannuronate and gelatin sponge. We managed to treat these two cases of mediastinal BAA with short neck by a combination of TAE and aorta stent. The average duration in the hospital after operation was 11.7 days for seven reviewed cases in **Table 1** as opposed to our two cases that were hospitalized for 19 days.

Our two case studies that we had presented here add further evidence to confirm the efficacy and safety of this approach in the man-

agement of mediastinal BAA. The endovascular method highlighted in literature is the optimal treatment for patients with suitable anatomic features. As shown in our case reports, we determined that patients with BAA and short neck length benefited most from the placement of aortic stent graft to exclude the aneurysm by closing the feeding vessels. It is also important to close the outflow arteries arising from the aneurysmal sac to prevent retrograde filling and subsequent risk of rupture of the revascularized BAA.

Conclusion

We have successfully demonstrated that mediastinal BAA can be treated with existing endovascular techniques. It is important that treatment is prompt even without any significant symptoms, as mediastinal BAA is potentially life threatening. We have shown that using a combined approach with aortic stent-graft placement and coil embolization, this benefited patients suffering from BAAs with short neck length. We believe that further follow-ups on these existing cases are necessary to ensure long-term treatment effectiveness. In order to better gauge the treatment efficacy, more case studies will have to be repeated for patients with similar anatomic features.

Acknowledgements

We thank the staff of Ningbo Medical University Affiliated Li Huili Hospital who aided in the study. We are also grateful for the two participants who provided consent for this case report.

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Hongcheng Wu, Department of Respiratory Medicine, Ningbo Medical University Affiliated Li Huili Hospital, Ningbo 315040, Zhejiang, China. E-mail: smj800722@aliyun.com

References

- [1] Mizuguchi S, Inoue K, Kida A, Isota M, Hige K, Aoyama T and Ishikawa T. Ruptured bronchial artery aneurysm associated with bronchiectasis: a case report. *Ann Thorac Cardiovasc Surg* 2009; 15: 115-118.
- [2] Hall RJ, Miller GA and Kerr IH. Ruptured bronchial artery aneurysm mimicking aortic dissection. *Br Heart J* 1977; 39: 909-910.
- [3] Tanaka K, Ihaya A, Horiuchi T, Morioka K, Kimura T, Uesaka T, Sasaki M, Uchinami M, Tsuda T and Yamada N. Giant mediastinal bronchial artery aneurysm mimicking benign esophageal tumor: a case report and review of 26 cases from literature. *J Vasc Surg* 2003; 38: 1125-1129.
- [4] Kasashima F, Endo M, Kosugi I, Matsumoto Y, Abe Y, Sasaki H, Sanada J and Matsui O. Mediastinal bronchial artery aneurysm treated with a stent-graft. *J Endovasc Ther* 2003; 10: 381-385.
- [5] Sakai T, Razavi MK, Semba CP, Kee ST, Sze DY and Dake MD. Percutaneous treatment of bronchial artery aneurysm with use of transcatheter coil embolization and thoracic aortic stent-graft placement. *J Vasc Interv Radiol* 1998; 9: 1025-1028.
- [6] Vosse BAH, van Belle AF, de Vries GJ and Das M. Hemomediastinum due to spontaneous rupture of a mediastinal bronchial artery aneurysm-A rare cause of thoracic pain. *Respir Med Case Rep* 2014; 12: 27-29.
- [7] Pugnale M, Portier F, Lamarre A, Halkic N, Riis H-B, Wicky S, Schnyder P and Denys A. Hemomediastinum caused by rupture of a bronchial artery aneurysm: successful treatment by embolization with N-butyl-2-cyanoacrylate. *J Vasc Interv Radiol* 2001; 12: 1351-1352.
- [8] Wilson SR, Winger DI and Katz DS. CT visualization of mediastinal bronchial artery aneurysm. *Am J Roentgenol* 2006; 187: W544-W545.
- [9] Abet D and Pietri J. [Ruptured bronchial artery aneurysm simulating dissection of the aorta in a patient with bronchopulmonary sequestration (author's transl)]. *J Chir (Paris)* 1981; 118: 743-746.
- [10] Sancho C, Dominguez J, Escalante E, Hernandez E, Cairals M and Martinez X. Embolization of an anomalous bronchial artery aneurysm in a patient with agenesis of the left pulmonary artery. *J Vasc Interv Radiol* 1999; 10: 1122-1126.
- [11] Shaer AH and Bashist B. Computed tomography of bronchial artery aneurysm with erosion into the esophagus. *J Comput Assist Tomogr* 1989; 13: 1069-1071.
- [12] Hoffmann V, Ysebaert D, De Schepper A, Colpaert C and Jorens P. Acute superior vena cava obstruction after rupture of a bronchial artery aneurysm. *Chest* 1996; 110: 1356-1358.
- [13] Cearlock JR, Fontaine AB, Urbaneja A and Spigos DG. Endovascular treatment of a posttraumatic bronchial artery pseudoaneurysm. *J Vasc Interv Radiol* 1995; 6: 495-496.

Treating BAA with aortic stent and embolization

- [14] Ishizaki N, Shimokawa S, Tanaka K, Taira A, Onohara S, Tabata M and Sakoda K. Ruptured bronchial artery aneurysm associated with pleural telangiectasis and tortuous portal obstruction: report of a case. *Surg Today* 1995; 25: 852-854.
- [15] Chantepie A, Robert M, Pelletier J, Gold F, Mercier C, Lacombe A and Laugier J. [Mycotic aneurysm of bronchial artery. Apropos of a case in an infant]. *Chir Pediatr* 1979; 21: 407-410.
- [16] Kalangos A, Khatchatourian G, Panos A and Faidutti B. Ruptured mediastinal bronchial artery aneurysm: a dilemma of diagnosis and therapeutic approach. *J Thorac Cardiovasc Surg* 1997; 114: 853-856.
- [17] Maisawa K and Koh E. [Ruptured bronchial artery aneurysm with left hemothorax: report of a case]. *Kyobu Geka* 2012; 65: 419-422.
- [18] Karmy-Jones R, Hastreiter D and Burdick T. Hemothorax complicating bronchial artery aneurysm. *Can Respir J* 2005; 12: 279-281.
- [19] Guzzardi G, Cerini P, Fossaceca R, Commodo M, Micalizzi E and Carriero A. Endovascular treatment of bronchial artery aneurysm with aortic stent-graft placement and coil embolization. *Ann Vasc Surg* 2012; 26: 1013-e1015.
- [20] Yanagihara K, Ueno Y, Kobayashi T, Isobe J and Itoh M. Bronchial artery aneurysm. *Ann Thorac Surg* 1999; 67: 854-855.
- [21] Takahashi Y, Tsutsumi Y, Monta O, Kohshi K, Ohashi H, Shimamura K and Osuga K. Stent grafting for giant bronchial artery aneurysm disguised as an aneurysm with multiple feeding arteries. *Ann Thorac Surg* 2010; 89: 1652-1654.
- [22] Sanchez E, Alados P, Zurera L, Canis M, Muñoz I, Casares J and Eguaras MG. Bronchial artery aneurysm treated with aortic stent graft and fibrin sealant. *Ann Thorac Surg* 2007; 83: 693-695.
- [23] Hu CX, Huang S, Xu ZW, Chen W, Huang JS And Fu Z. Combination of aortic stent-graft and arterial embolization for ruptured bronchial artery aneurysm. *Ann Thorac Surg* 2011; 92: e19-e21.
- [24] Bouayad M, Bagan P, Brian E, Benabdesselam A and Couffinhal JC. [Mediastinal bronchial artery aneurysm: treatment with stent-graft]. *J Mal Vasc* 2011; 36: 45-49.