

Endobronchial hemangioma: an unusual case. Case report

Hemangioma endobronquial: un caso inusual. Presentación de caso

Juan P. Manrique-Salas[1]
Ricardo Méndez-Maho[1]
Hugo C. Gutiérrez-González[1]
Julita del Socorro Orozco-Vázquez[2]
Mario Lemus-Velázquez[3]

https://doi.org/10.53903/01212095.231

Key words (MeSH)

Bronchial diseases
Hemangioma, capillary
Radiology, interventional
Embolization, therapeutic

Palabras clave (DeCS)

Enfermedades bronquiales
Hemangioma capilar
Radiología intervencionista
Embolización terapéutica

Summary

Tracheobronchial tree capillary hemangioma is extremely rare in adults, with hemoptysis being one of the most serious forms of presentation. Surgery has been the treatment of choice, although it implies high rates of morbidity and mortality, which has led to the search for other therapeutic alternatives. The case of a 54-year-old man with a hemangioma in the right main bronchus with extension to the carina and trachea is presented. Resection and sclerotherapy by fiberoptic bronchoscopy was attempted, which was unsuccessful due to abundant bleeding. Therefore, supraselective embolization was performed, and later perform surgical resection.

Resumen

Los hemangiomas capilares del árbol traqueobronquial son extremadamente raros en adultos, y la hemoptisis es una de las formas más graves de presentación. La cirugía ha sido el tratamiento de elección, aunque implica altas tasas de morbilidad y mortalidad, lo que ha llevado a la búsqueda de otras alternativas terapéuticas. Se presenta el caso de un hombre de 54 años de edad, quien cursa con un hemangioma en el bronquio principal derecho con extensión a carina y tráquea. La resección y escleroterapia por fibrobroncoscopia fue fallida, debido a sangrado abundante; por ende, se realizó embolización supraselectiva y, posteriormente, resección quirúrgica.

Introduction

Primary endobronchial tumors are rare, representing 0.1-0.4% of airway tumors; they are usually malignant in adults and benign in children (1).

Lobular capillary hemangioma is a benign tumor that commonly occurs in skin and mucosa of the upper airway, characterized by rapid growth and a friable surface (2).

The etiology of capillary hemangiomas has not been completely defined. However, there are postulates that associate this pathology with hormonal changes, trauma, viral or bacterial infections, production of angiogenic factors and clonal cytogenetic deletions (3).

In adults, the most frequent causes of cough and hemoptysis are malignant tumors, infectious diseases (pneumonia, chronic bronchitis and tuberculosis), cardiovascular disorders and other inflammatory diseases (4).

There is no consensus on the best treatment of endobronchial hemangiomas, perhaps because of their low incidence, but some therapeutic options have been described; among them, flexible or rigid bronchoscopy with different modalities such as cryoablation, electrocautery, photocautery and surgical management (5).

Case report

A 54-year-old man with a history of chronic obstructive pulmonary disease presented with hemoptysis. A multiphase computed axial tomography (CT) of the thorax showed a lobulated lesion emerging from the right main bronchus and generating total occlusion, the lesion extended towards the middle lobe, carina and distal trachea; atelectasis of the ipsilateral middle and lower lobe was also observed (figure 1), and early arterial enhancement in the phase with contrast medium (figure 2).

In view of this finding, bronchoscopy was performed to take a biopsy and an attempt was made to perform excision, which failed due to profuse bleeding; it was suspended, bleeding was controlled with argon plasma and the biopsy was performed, with the result of capillary hemangioma. Embolization was performed prior to surgical treatment to reduce the risk of hemorrhage. Chest CT angiography was performed as part of the planning of the procedure; it showed a mass with lobulated borders emerging from the right main bronchus with intermediate signal in T1 and high signal in T2, with avid enhancement with contrast medium (Figure 3).

[1]Resident physician specializing in Diagnostic and Therapeutic Imaging. National Autonomous University of Mexico. Department of Diagnostic and Therapeutic Imaging, Hospital Centro Médico Nacional 20 de Noviembre, Mexico City, Mexico.

[2]Specialist in Diagnostic and Therapeutic Imaging. National Autonomous University of Mexico. Head of the Department of Diagnostic and Therapeutic Imaging, Hospital Centro Médico Nacional 20 de Noviembre, Mexico City, Mexico.

[3]Interventional Radiologist. General Hospital of Mexico. National Autonomous University of Mexico. Department of Diagnostic and Therapeutic Imaging, Hospital Centro Médico Nacional 20 de Noviembre, Mexico City, Mexico.

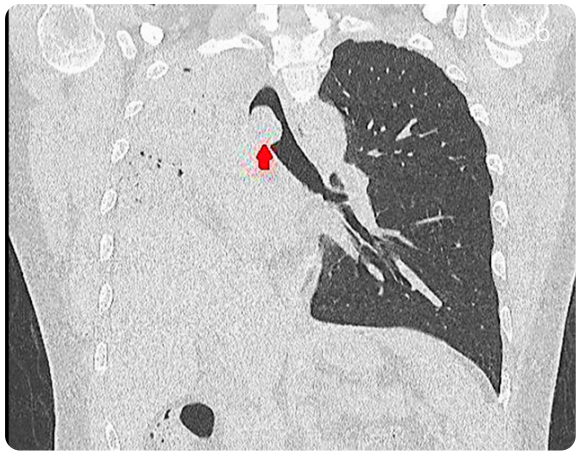


Figure 1. Simple chest CT in coronal reconstruction, in pulmonary parenchyma window. Lobulated mass in the right main bronchus with total occlusion, which extends towards the middle lobe, carina and distal trachea (arrow); ipsilateral middle and lower lobe atelectasis, and mediastinal displacement.



Figure 2. Chest CT with contrast medium in arterial phase in coronal reconstruction, in mediastinal window. Lobulated mass in the right main bronchus, with moderate homogeneous enhancement to the passage of contrast medium (arrow).

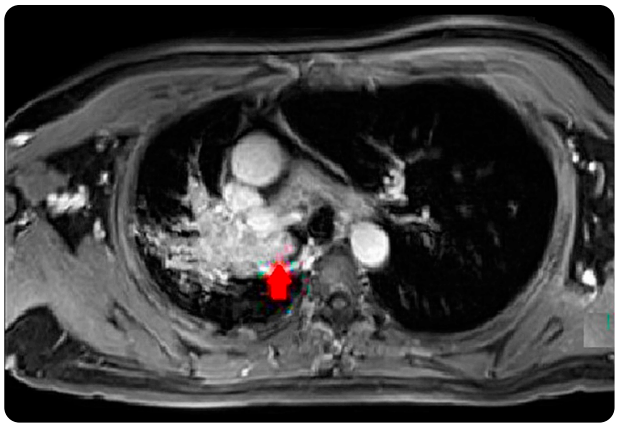


Figure 3. MRI in axial acquisition, T1-weighted with intravenous contrast medium. Mass with lobulated borders emerging in the right main bronchus with avid enhancement on passage of contrast medium.

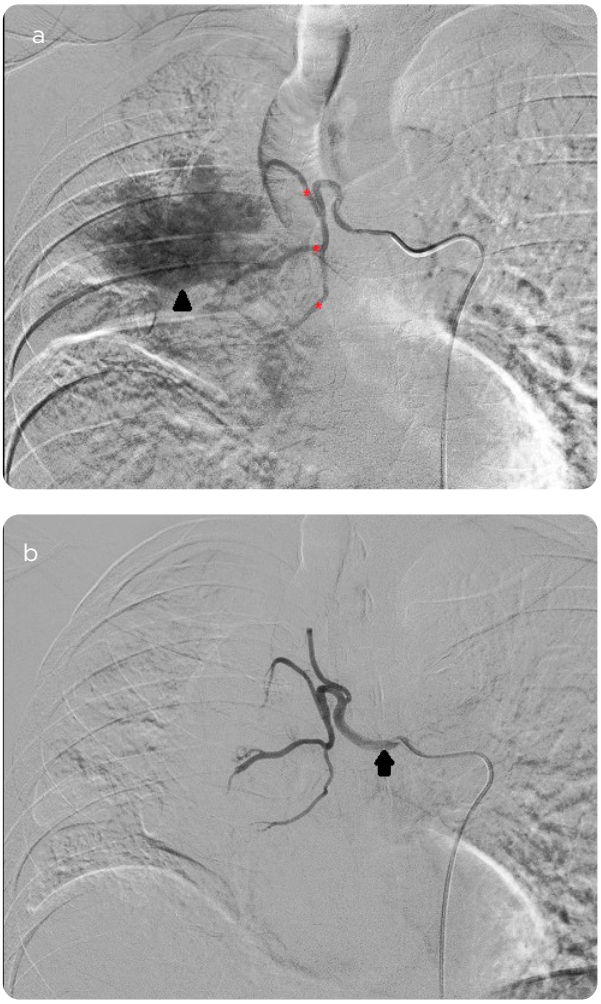


Figure 4. Angiography with magnification and subtraction in anteroposterior projection. a) Pre-embolization and b) post-embolization. There is evidence of vessel trifurcation (*) arising at the level of the thoracic descending aorta (arrow), and sinuous trajectory leading to the site of malformation (arrowhead). Embolization with polyvinyl alcohol microspheres until achieving absence of lesion enhancement (b).

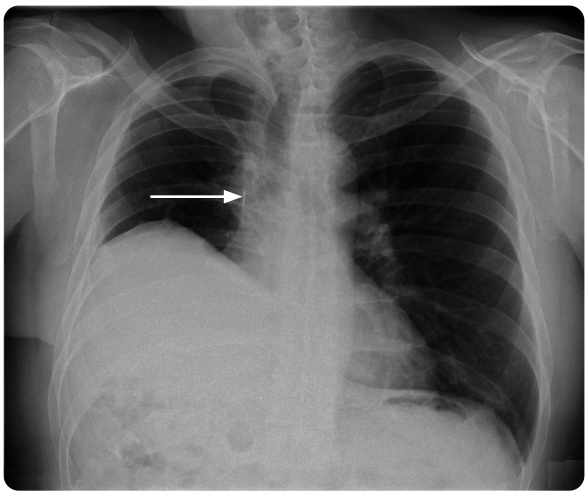


Figure 5. Chest X-ray with adequate right pulmonary expansion, right main bronchus without occupation, elevation of the right hemidiaphragm due to pulmonary segmentectomy, linear right hilar radiopaque image is observed in relation to surgical material (arrow).

Hemangiomas have a high signal secondary to their water content on magnetic resonance imaging (MRI) in T2-weighted sequences. Hemangiomas are visualized with high signal on T2-weighted images, while tumors show some loss of signal intensity. On contrast-enhanced T1-weighted MRI they have a slow centripetal pattern of peripheral enhancement, a finding that is more specific for the diagnosis of hemangioma. Less frequently, hemangiomas may have atypical features such as heterogeneity, rapid enhancement, calcification, thrombosis, hemorrhage, fluid-liquid levels and fibrosis (6).

For the embolization procedure, thoracic aorta angiography is performed using the Seldinger technique with entry into the right common femoral artery with vascular introducer 6 Fr. With fluoroscopic guidance, the hydrophilic guide and Pig-Tail 5 Fr catheter are introduced. In this arteriography the arterial fistulous trajectory dependent on the thoracic descending aorta is visualized by its right border in T6-T7, with vessel trifurcation and sinuous trajectory that ends in the malformation site, so the Pig-Tail catheter is removed and the 5 Fr curved Cobra catheter is introduced. The dimensions of the nutrient vessel of the hemangioma are 4.6 mm proximally and 4.2 mm distally. Additionally, adjacent atelectasis and mediastinal displacement are observed. Embolization was performed with polyvinyl alcohol microspheres (300-500 microns) diluted with iodinated contrast medium until there was no opacification of the lesion (figure 4). Bronchoscopy was performed to remove the lesion and later segmentectomy of the medial segment of the middle lobe was performed.

Chest X-ray after the procedure showed adequate pulmonary expansion with elevation of the right hemidiaphragm due to segmentectomy (figure 5).

Discussion

Airway capillary hemangiomas respond well to bronchoscopic intervention (7). DeKeratry reported a case with a favorable outcome using flexible bronchoscopic intervention with argon plasma coagulation (APC); he argued that the established safety of APC makes it an ideal tool for treating this type of lesion (7). However, in the case presented, a repeat procedure was required to control recurrent hemoptysis after the initial intervention. Zambudio et al. described the first arteriographic embolization to treat a capillary hemangioma located in the proximal trachea (8, 9). There are four cases described in the literature with good therapeutic outcome of similar lesions, treated by excisional forceps biopsy procedure with adequate hemostasis, minimal morbidity and no mortality (9). The short-term success rate of bronchial arterial embolization for vascular lesions causing hemoptysis is about 80-90% (10).

Endobronchial hemangiomas can cause massive bleeding. Therefore, CT and/or contrast angiography should be considered before biopsy, in case similar findings are observed by bronchoscopy. Endobronchial hemangiomas treated with arterial embolization may in their evolution present with residual lesions and/or recanalization of these, which may warrant consideration of further treatment, including surgery (11).

Surgery has been the treatment of choice, although it involves high morbidity and mortality rates, especially in emergency situations such as massive hemoptysis, so other therapeutic alternatives have been sought. There is little experience with embolization by interventional radiology when hemoptysis is of tracheal origin, partly because of the infrequency of this pathology; however, the foundations for this have been laid with the development of embolization for other bronchopulmonary pathologies (9).

The long-term success of embolization depends not only on the quality of arterial occlusion, but also on the efficacy of medical treatment. The prognosis of severe hemoptysis can be improved with arterial embolization (12).

In the case documented here, embolization was a safe alternative prior to the procedure with good patient response. The risk of bleeding and complications inherent to the surgical procedure were minimized.

References

1. Grillo Hermes C, Mathisen D. Primary tracheal tumors: treatment and results. Ann Thor Surg. 1990;49(1): 69-77.
2. Dabó H, Gomes R, Teixeira N, Teixeira G, Fernandes G, Magalhães A. Tracheal lobular capillary hemangioma treated with laser photocoagulation. J Bras Pneumol. 2016;42(1):72-3. https://doi.org/10.1590/S1806-37562016000000217
3. Chen E, Yu X, Zhang R, Wang P, Ying K. A large tracheal capillary hemangioma treated with interventional bronchoscopy. Respiratory Medicine CME. 2011;4(2):60-1. https://doi.org/10.1016/j.rmedc.2010.10.002
4. Chawla AK, Chaudhary G, Aggarwal M, Chawla MK, Chawla RK. Endobronchial capillary hemangioma a rare cause of hemoptysis. Lung India: oficial organ of Indian Chest Society. 2020;37(5):445-8. https://doi.org/10.4103/lungindia.lungindia_508_19
5. Chawla M, Stone C, Simoff MJ. Lobular capillary hemangioma of the trachea: the second case. J Bronchology Interv Pulmonol. 2010;17(3):238-40. https://doi.org/10.1097/LBR.0b013e3181e65117
6. Vilanova JC, Barceló J, Smirniotopoulos J, Pérez-Andrés R, Villalón M, Miró J, et al. Hemangioma from Head to Toe: MR imaging with pathologic correlation. RadioGraphics. 2004;24(2):367-85. https://doi.org.10.1148/rg.242035079
7. DeKeratry DR. Argon plasma coagulation for endobronchial hemangioma: a new treatment option for a rare cause of hemoptysis. J Bronchol. 2004;11:254-6.
8. Rose AS, Mathur PN. Endobronchial capillary hemangioma: case report and review of the literature. Respiration. 2008;76(2):221-4. https://doi.org/10.1159/000099008
9. Zambudio AR, Calvo MJ, Lanzas JT, García J, Parrilla P. Massive hemoptysis caused by tracheal hemangioma treated with interventional radiology. Ann Thorac Surg. 2003;75:1302-4. https://doi.org/10.1016/s0003-4975(02)04656-8
10. Yoshida T, Kato M, Takeda N. A case of primary racemose hemangioma of the bronchial artery that disappeared after bronchial artery embolization. J. Jpn Soc. Respir. Endosc. 2014;36(3):256-60.
11. Imai S, Kasai H, Sugiura T, Nagata J, Toyoda T, Shiohira S, et al. A case of primary racemose hemangioma with endobronchial lesions demonstrating recurrent hemoptysis initially treated with bronchial arterial embolization. Respir Med Case Rep. 2022;40:101762. https://doi.org/10.1016/j.rmcr.2022.101762
12. Rémy J, Arnaud A, Fardou H, Giraud R, Voisin C. Treatment of hemoptysis by embolization of bronchial arteries. Radiology. 1997:122(1):33-7.

Correspondence

Juan P. Manrique-Salas
Avenida Félix Cuevas 540,
Col del Valle Sur, Benito Juárez, 03229
Ciudad de México, México jpmansa@gmail.com

Received for evaluation: March 20, 2023
Accepted for publication: May 20, 2023