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Two Bronchial Artery Aneurysms in Cystic Fibrosis

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Abstract

Bronchial artery aneurysm (BAA) is a rare finding often found in patients with underlying diseases such as cystic fibrosis. We present a case of two BAAs in a 55-year-old male patient with a diagnosis of cystic fibrosis, which had been misdiagnosed as lymph nodes in a prior non-contrast computed tomography study, as enlarged lymph nodes are much more commonly seen than BAAs. High mortality is associated with BAA ruptures; thus, accurate and timely diagnosis along with treatment is especially crucial.

Keywords: Bronchial artery aneurysm, CT scan, cystic fibrosis

Introduction

Bronchial arterial aneurysm (BAA) refers to aneurysmal dilatation involving any segment of the bronchial artery. It is a rare entity and has been found in <1% of the population who undergo selective bronchial arterial angiograms.[1] The pathogenesis of them is not well known. Most of the cases have underlying predisposing pulmonary or systemic diseases such as bronchiectasis, silicosis, lung cancer, recurrent infection, thoracic trauma, or Osler–Weber–Rendu syndrome.[2,3] They are also found in patients with atherosclerosis and individuals with predisposing congenital conditions such as cystic fibrosis.[4] We present a case of two BAAs in a 55-year-old male patient with the diagnosis of cystic fibrosis, which had been misdiagnosed as lymph nodes in a prior non-contrast computed tomography (CT) study.

Case Presentation

Clinical Findings

A 55-year-old Caucasian male with a history of cystic fibrosis (CF), diabetes mellitus, and erectile dysfunction presented for evaluation of dysphagia. He reported that he had mild dysphagia symptoms for the past decade, which progressed to moderate severity recently. He used to have intermittent episodes of dysphagia to solid foods, currently occurring about once daily. He sometimes had to force the material down with liquids or regurgitate it for relief. The patient did not have trouble initiating the swallow nor did he choke or aspirate. He had no family history of esophageal or gastric cancer. The patient's vital signs were normal and stable, and his physical exam was insignificant.

Imaging Findings

A non-contrast CT study performed 10 months ago revealed bilateral diffuse bronchial wall thickening, cylindrical bronchiectasis, or mucus plugging in the bronchioles causing bronchial occlusion along with the near complete fatty replacement of the pancreas, which are consistent with the patient's known diagnosis of CF and several enlarged mediastinal lymph nodes.

The chest CT with contrast showed similar findings to the prior study with two BAAs measuring 1.2 and 1.6 cm, which were misdiagnosed as enlarged lymph nodes due to the non-contrast nature of that study (Figures 1 and 2).

Discussion

BAAs are very rare. They can be in mediastinal/juxta aortic or intrapulmonary location. Most of the cases are asymptomatic. If they are big enough, they might present as large mediastinal mass and can cause dysphagia or compression findings to adjacent structures such as superior vena cava syndrome.[5] Our case also presented with dysphagia symptoms. BAA that originates from the inferior segment of the aortic arch may mimic aortic aneurysms.[6] The most dangerous complication is catastrophic hemorrhage due to rupture.

BAA can be diagnosed by CT, MR angiography, or catheter angiography.[1,7-9] In the vast majority of cases requiring chest CT, the use of contrast media is not required for an accurate diagnosis. The most common indications for CT, such as interstitial lung disease, COPD, pulmonary nodule, airway diseases, and lung cancer screening, do not require the use of any contrast media. However, for the assessment of vascular diseases

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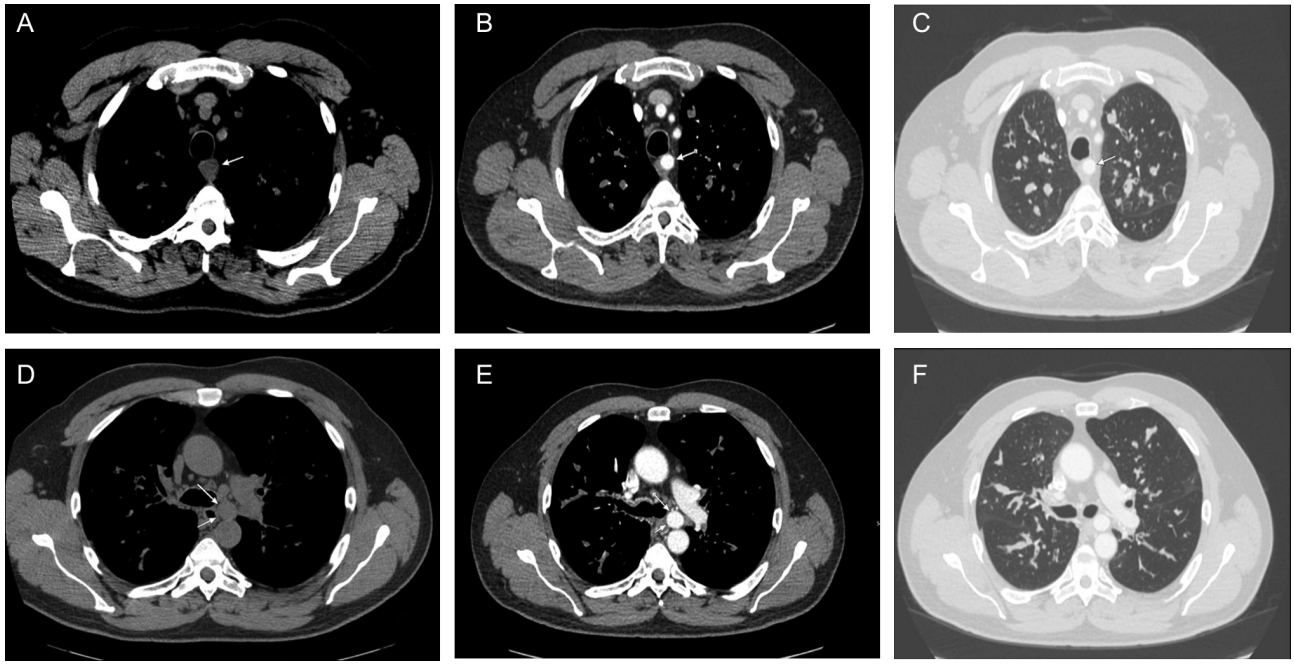

Figure 1. A 55-year-old man with a history of cystic fibrosis (CF). Axial CT images of the chest at mid-tracheal level (A, B, C) and at the level of the Carina (D, E, F). (A and D) Non-contrast enhanced computed tomography (NCECT) mediastinal window images demonstrate two rounded lesions: one behind the trachea and the other anterior to the descending thoracic aorta in the aorto-pulmonary window (arrows). They are thought to be enlarged lymph nodes. (B and E) Contrast enhanced computed tomography (CECT) mediastinal window images demonstrate enhancement of the previously mentioned lesions (arrows) consistent with vascular structures (bronchial artery aneurysms) without contrast leak. (C and F) CECT lung window images demonstrate background abnormalities of CF with bronchial wall thickening, endobronchial mucus plugging, and bronchiectatic changes.

such as aneurysm, dissection, and vascular tumor invasion or pulmonary embolism, CT needs to be performed with intravenous contrast to delineate the vessel lumen and differentiate vascular pathologies from other mediastinal structures such as lymph nodes.

In our case, the BAAs have been misdiagnosed as enlarged lymph nodes in the first non-contrast CT study as the enlarged lymph nodes are much more common than BAA.

CF is a disease with an autosomal recessive pattern with incidence of around 1:2000-3500 live births. It is caused by the mutation in the cystic fibrosis transmembrane conductance regulator (CFTR) gene found on chromosome 7.[10] It interrupts the exocrine function of the lungs, pancreas, liver, small intestines, sweat glands, and the male genital system and thus causes progressive incapacity and multisystem failure.[10] Pulmonary involvement of cystic fibrosis is commonly described and therefore the best studied, which are central and upper lobe

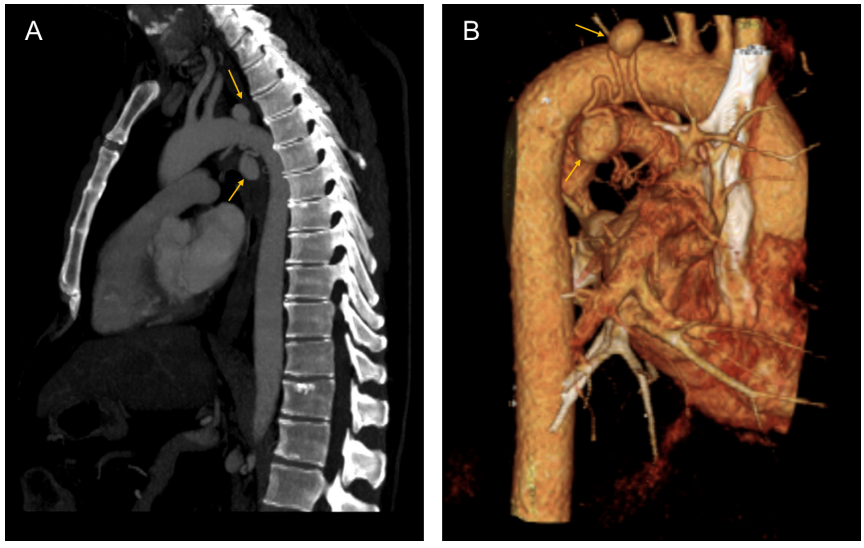

Figure 2. CECT oblique sagittal maximum intensity projection image (A) and 3D volume rendering technique image (B) of the same patient demonstrate the bronchial artery aneurysms (yellow arrows) along the course of the bronchial arteries originating from the thoracic aorta.

predominant bronchiectasis, bronchial wall thickening, centrilobular and tree-in-bud opacities, branching nodules, finger in glove appearance due to mucus plugging within the bronchi and air trapping.

BAAs can be seen in contrast-enhanced studies. It is important to be aware of potential vascular findings such as BAAs. Therefore, it is very important for the radiologist and clinicians to be cognizant of the limitations of non-contrast studies.

When a BAA diagnosis is confirmed, it is important to treat it as soon as possible as there is a high mortality rate associated with BAA rupture.[11] The treatment options are transcatheter embolization, covered stent placement, or surgery.[2,4,12-15]

Transcatheter embolization is accepted as first-line management due to its low invasive nature. It is, however, difficult to achieve complete embolization when the vascular segment between the aneurysm and aorta is short. There is also the possibility of revascularization of BAA by collateral vessels, incomplete embolization, and arterial re-canalization with this technique. Hence, stent placements have been noted in the past years as an alternate treatment option. By sealing the opening site of the feeding artery, this technique makes it possible to completely isolate the blood flow into the aneurysm. This technique has been used in combination with transcatheter embolization to achieve better results as well. Lastly, in the surgical approach, resection of BAA can be performed through thoracotomy. While this approach makes it possible to remove a lesion completely, it also is an invasive procedure that is reserved mostly for patients who are contraindicative for endovascular treatment options.[11]

Informed Consent: Written informed consent was obtained from the patients who participated in this study.

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References

1. Zhang LJ, Yang GF, Lu GM. Noninvasive diagnosis of bronchial artery aneurysm using dual-source computed tomography angiography. *Acta Radiol*. 2008;49(3):291-294. [CrossRef]
2. Yoon W, Kim JK, Kim YH, Chung TW, Kang HK. Bronchial and nonbronchial systemic artery embolization for life-threatening hemoptysis: a comprehensive review. *RadioGraphics*. 2002;22(6):1395-1409. [CrossRef]
3. Adachi K, Tarukawa T. Mediastinal bronchial artery aneurysm associated with bronchiectasis; report of a case. *Kyobu Geka*. 2018;71(5):396-399.
4. Kalva SP, Wicky S. Mediastinal bronchial artery aneurysms: endovascular therapy in two patients. *Catheter Cardiovasc Interv*. 2006;68(6):858-861. [CrossRef]
5. Wilson SR, Winger DI, Katz DS. CT visualization of mediastinal bronchial artery aneurysm. *Am J Roentgenol*. 2006;187(5):W544-W545. [CrossRef]
6. Vernhet H, Bousquet C, Jean B, et al. Bronchial aneurysms mimicking aortic aneurysms: endovascular treatment in two patients. *Cardiovasc Intervent Radiol*. 1999;22(3):254-257. [CrossRef]
7. Remy-Jardin M, Remy J, Ramon P, Fellous G. Mediastinal bronchial artery aneurysm: dynamic computed tomography appearance. *Cardiovasc Intervent Radiol*. 1991;14(2):118-120. [CrossRef]
8. Shaer AH, Bashist B. Computed tomography of bronchial artery aneurysm with erosion into the esophagus. *J Comput Assist Tomogr*. 1989;13(6):1069-1071. [CrossRef]
9. Bak SH, Han H. Diagnosis of bronchial artery aneurysm by computed tomography: a case report. *Rad Case Rep*. 2017;12(3):455-459. [CrossRef]
10. Maffessanti M, Polverosi R, Dalpiaz G, et al. *Diffuse Lung Diseases, Clinical Features, Pathology, HRCT*. Berlin: Springer Verlag; 2006.
11. Yanagihara K, Ueno Y, Kobayashi T, Isobe J, Itoh M. Bronchial artery aneurysm. *Ann Thorac Surg*. 1999;67(3):854-855. [CrossRef]
12. Di X, Ji DH, Chen Y, et al. Endovascular treatment of ectopic bronchial artery aneurysm with brachiocephalic artery stent placement and coil embolization: a case report and literature review. *Medicine*. 2016;95(35):e4461. [CrossRef]
13. Li Y, Gu GC, Liu B, et al. Endovascular treatment of multiple bronchial artery aneurysms with prominent fistula to pulmonary artery in a patient with interstitial lung disease: a case report and literature review. *Vasc Endovasc Surg*. 2019;53(6):492-496. [CrossRef]
14. Matsumoto T, Uchida T, Ono T, et al. Bronchial artery aneurysm treated using aortic stent graft alone: a case report. *Ann Vasc Dis*. 2017;10(2):152-154. [CrossRef]
15. Lü PH, Wang LF, Su YS, et al. Endovascular therapy of bronchial artery aneurysm: five cases with six aneurysms. *Cardiovasc Intervent Radiol*. 2011;34(3):508-512. [CrossRef]