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Case Report: A Rare Case of Coronary-Bronchial Fistula Associated with a Large Lung Bullae and Bronchiectasis Presenting as Dyspnea

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Abstract: Coronary artery fistulas (CAF) are rare but hemodynamically significant anomalies. Although asymptomatic, they can be associated with several cardiorespiratory conditions. Coronary to bronchial fistulas (CBF) account for 0.5% to 0.61% of coronary artery fistulas, with fistulas arising from the right coronary artery being exceedingly rare. These fistulas are known to be associated with bronchiectasis but not lung bullae. The following paper reports a rare case of a coronary to bronchial fistula associated to bronchiectasis and lung bullae. The patient presented for dyspnea and was found to have a large lung bullae, bronchiectasis and a coronary to bronchial artery fistula arising from the right coronary artery and terminating into the left bronchial artery. The CBF was successfully managed first with percutaneous microcoil embolization then the bullae was resected thoracoscopically three days later. However, more case reports are mandatory in order to further understand the etiology and pathophysiology of these fistulas, elucidate their relationship to other pathologies such as bronchiectasis and lung bullae and determine the optimal therapeutic measures.

Keywords: Coronary Bronchial Artery Fistula, Bronchiectasis, Lung Bullae, Microcoil Embolization

1. Introduction

Coronary artery fistulas (CAF) Accounts for 0.3% of congenital heart diseases. They are rare anomalies of congenital or acquired origins [1], often incidentally discovered on invasive angiography given their asymptomatic nature, especially during the first 2 decades of life [2-3]; however, these fistulas are the most common coronary artery anomalies that can alter coronary hemodynamic parameters [4], and are associated with angina, myocardial infarctions, heart failure, arrhythmias, and infective endocarditis [1-3]. Their prevalence ranges between 0.002% to 0.4% on coronary angiography [1], however computed tomographic angiography (CTA) reveals a prevalence close to 0.9% [5]. Of the CAF types, coronary artery to bronchial artery fistulas (CBF) are the rarest with prevalence rates of 0.5% on coronary angiography and 0.61% on CTA [1]. Furthermore, though CAFs arise from the RCA in 50% to 55% of cases, CBFs commonly arise from the left circumflex artery [1, 6] making an origin from the RCA an exceedingly rare occurrence. The following case describes a CBF arising from the RCA to the left bronchial artery in a

patient with a large emphysematous bullae. The fistula was successfully managed with percutaneous microcoil embolization while the bulla was resected few days later.

2. Case Report

A 66-year-old lady presented to our hospital three months ago for shortness of breath of 15 days duration. She is known to have a congestive heart failure with diastolic dysfunction, with septo-apical dysrhythmia and an ejection fraction of 40% managed with aspirin and bisoprolol as well as chronic obstructive pulmonary disease managed with tiotropium bromide & olodaterol, beclomethasone dipropionate and on demand ipratropium bromide and 1L oxygen via nasal canula. Furthermore, the patient is an ex-smoker with a 140 pack-year history. At presentation, the patient reported exertional dyspnea with associated pleuritic chest pain and physical exam was significant for bilateral diffuse wheezing on auscultation. Electrocardiogram revealed a normal sinus rhythm with no evidence of ST changes suggestive of myocardial infarction. A CT scan was done that revealed a large emphysematous bulla measuring up to 14cm in the basilar segments of the left lower lobe as well as bilateral diffuse panlobular emphysematous changes, varicose bronchiectasis in the posterior segments of both upper lobes and moderate calcific atherosclerosis of the thoracic aorta & coronary arteries. An echocardiogram was done that revealed normal systolic function with delayed relaxation, an ejection fraction of 59% and mild mitral regurgitation. Cardiac catheterization was done using JL4 and JR5 5F catheters via the right radial artery approach. Catheterization revealed a large right coronary to bronchial artery fistula. (figure 1, and figure 2)

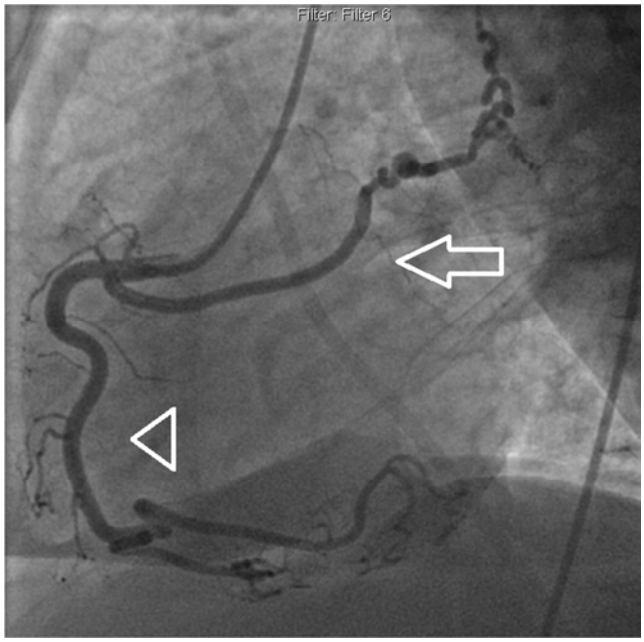

Figure 1. *Fistula (arrow) arising from the proximal segment of right coronary artery (arrowhead) and terminating in the left bronchial artery with no involvement of the pulmonary arteries.*

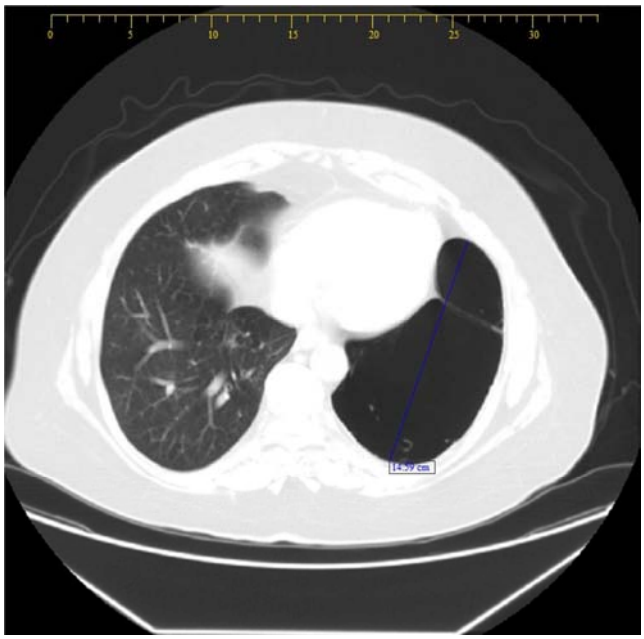

Figure 2. *CT Scan demonstrating emphysematous changes and bronchiectasis present in the left lower lobe.*

The RCA ostium was subsequently catheterized using an AL1 guide catheter and a 0.021 Progreat microcatheter was advanced to the distal segment of the fistula. The fistula was embolized using two Micronester 6mm coils & and one Micronester 5mm coil. Test injection into the right RCA ostium revealed no communication between the RCA and left pulmonary tree. (figures 3, 4)

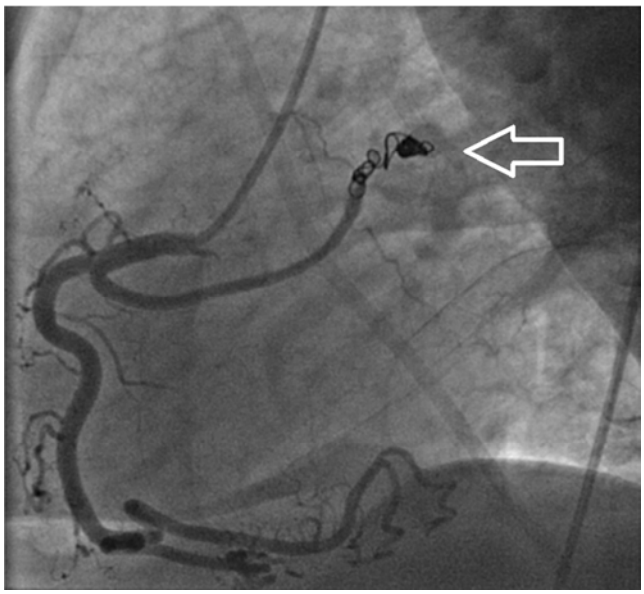

Figure 3. *Microcoil embolus (arrowhead) in the distal segment of the coronary to bronchial artery fistula.*

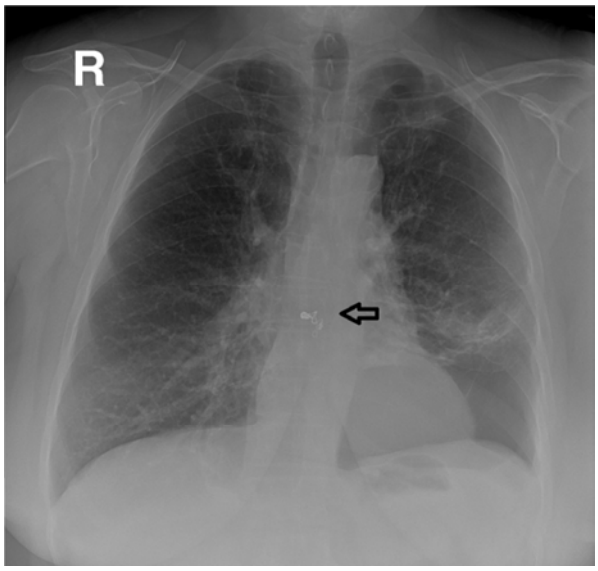

Figure 4. *Antero-posterior view chest x-ray following coil embolization and demonstration the left lower lobe bulla prior to surgical resection.*

The patient underwent thoracoscopic bullectomy 3 days following the embolization. had a smooth postoperative course and was discharged 6 days after surgery.

3. Discussion

Coronary artery fistulas are rare anomalies. Classically, the prevalence was estimated to be 0.002% on coronary angiography [7]. Further studies performed using CTA revealed an incidence closer to 0.9% [6]. Erdem and Ozbay determined that the total incidence of coronary artery anomalies is closer to be 3.9% on coronary angiography [6]. Coronary-bronchial fistulas make of the rarest classification of coronary artery fistulas with a prevalence of 0.5% at coronary angiography and 0.61% on CTA [1]. 90% of CAFs are of congenital origins secondary to failure of regression of myocardial sinusoids, as with coronary-cameral fistulas, or the persistence of primitive communications between coronary & mediastinal vessels [1]. Acquired CAFs are iatrogenic and occur secondary to prior coronary bypass graft surgery, coronary stent placement, irradiation of the chest, or chest trauma. Myocardial infarction and coronary vasculitis are also recognized as causes of coronary artery fistulas [1]. Clinically, coronary artery fistulas are usually asymptomatic [1-2]. Symptoms are unlikely to develop in the first two decades of life but may develop depending chiefly on the size of the fistula and the distal draining site [1]. In fact, drainage into systemic circulation creates a left-to-right shunt with right heart strain with subsequent development of pulmonary hypertension and high-output he. Accordingly to an article from the Netherlands, only 31 such fistulas were reported in a period from 2008-2013. The etiology of theses fistulas is uncertain. Said et al suggested the possibility of the reopening of a preexisting non functional congenital communications between the bronchial arteries. they proposed that two factors regulate the reopening and growth of the arterial communication: Desequilibrium of the pressure gradient between the two arteries and obstruction of the coronary artery [8]. Many case series have implied the possibility of a relationship between CBF and bronchiectasis in one or both lungs [9]. This association was observed in our patient since the CT scan showed bronchiectasis in both upper lobes. In addition she was an ex smoker and had a large emphysematous bulla measuring up to 14cm in the basilar segments of the left lower lobe as well as bilateral diffuse panlobular emphysematous changes. The bullae was resected after embolization of the CBF. No similar cases presenting simultaneously CBF and emphysematous bullae were reported in the literature and no correlation between these two pathologies was discussed previously.

Clinical presentation of patients with CBF depends on the degree of left to right shunt and the concomitant disease process in the patients. Said et al reported that chest pain was the most frequent symptom (63%) that was mainly due to a coronary steal phenomenon [8, 10], hemoptysis (26%), Dyspnea (19%). Asymptomatic disease occurred only in (19%) [10]. Our patient was admitted for exertional dyspnea and pleuritic chest pain. no hemoptysis nor chest pain were reported. However the respiratory symptoms may not be only due to the presence of the CBF but may also be related to the respiratory status of the patient and the presence of a large bullae in the left lower lobe.

Similar to coronary artery fistulas, symptomatic coronary bronchial fistulas can be managed surgically or with percutaneous embolization with microcoils or a detachable balloon. Though several reports establish that surgery is well-tolerated with minimal post-op complications, percutaneous embolization is favored to minimize the morbidity of general anesthesia and median sternotomy, given that the patient is a candidate for embolization. Conservative treatment may be an option depending on the case. Galli et al. discussed a case that was successfully managed with non-dihydropyridine calcium channel blockers & oral nitrates in conjunction to the patient's angiotensin II receptor blocker and oral aspirin [11]. Hackett & Hallidie-Smith reported a case of spontaneous thrombosis & resolution of a coronary artery fistula [12]. Since 1-2% of cases resolve spontaneously, symptomatic management with antiplatelet therapy & antibiotics and close follow up is feasible [1], however according to the American College of Cardiologists and the American Heart Association, interventional management with percutaneous closure or surgical ligation is a Class I recommendation for large fistulas regardless of symptoms or for symptomatic small to moderate-sized fistulas [13]. Complications following percutaneous embolization have been reported. Recanalization occurs at variable rates. In one study, recanalization did not occur at the 6 month and 1 year follow ups [14]. Other studies however reported a rate of 10% of cases regardless of the type of intervention [15-16]. Recanalization occurs within the first year, making follow-up evaluation imperative [17]. Other complications are also reported, including myocardial infarction [1, 18], arrythmia [1, 15-16], coil migration & coronary artery dissection [19], as well as coronary artery spasm, dissection & perforation

[1]. In our case, no complications related to the embolization was observed and the patient underwent a bullectomy three days post embolization and was discharged 6 days after surgery.

4. Conclusion

Coronary artery fistulas are generally asymptomatic, however they can alter significantly the coronary hemodynamic parameters. Though rare, recent series determined that the prevalence is higher than previously believed. Of the CAF types, coronary artery to bronchial artery fistulas (CBF) are the rarest with prevalence rates of 0.5% on coronary angiography and 0.61 on CTA. These fistulas can be congenital or acquired. When present, management with surgical ligation or percutaneous embolization is recommended. Though several reports ascertain that surgery is well-tolerated with an uneventful postoperative course, embolization seems to be the preferred modality when applicable to reduce morbidity. This paper reports a rare combination of CBF, bronchiectasis and lung bullae. A strong correlation have already been described between CBF and bronchiectasis but not lung bullae. Therefore, more case reports are mandatory in order to further understand the etiology and physiopathology of theses fistulas, elucidate their relationship with other diseases (such as bronchiectasis bullae and emphysema), and determine the optimal therapeutic measure.

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