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Coronary-Bronchial Artery Fistula Presenting as Angina: A Case Report with Review of literature

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Abstract

Coronary-bronchial fistula is an extremely rare coronary anomaly often found incidentally on coronary angiography. We report the case of a 53-year-female presenting with angina on exertion incidentally detected to be having CBAF as the cause of angina and managed successfully with optimal medical therapy.

Keywords: angina; angiogram; congenital anomaly; coronary artery anomaly; coronary-bronchial fistula (CBAF)

Case report

A 53-year-old woman presented to our hospital with a 1-month history of angina on exertion (NYHA-Class II). There was no history of breathlessness, syncope, and hemoptysis. No past history of hypertension/diabetes/ hypothyroidism/ chronic kidney disease/stroke was noted. It was noted that 2 days prior to her consultation in out patient department (OPD), she had one episode of typical angina while walking, which subsided on rest. She was overweight, with a BMI of 32. Her complete hemogram, creatinine, cardiac biomarkers, and electrolytes were within normal limits. Chest X-Ray was unremarkable (Figure 1), electrocardiogram revealed generalized T inversions (Figure 2). Transthoracic echocardiography showed normal scan, with normal left ventricle function and absence of any regional wall motion abnormality. As patient denied treadmill stress test due to low physical fitness, and as the suspicion of coronary artery disease based on history was high, she was subjected to invasive coronary angiogram. Her coronary angiogram showed normal epicardial coronaries. A large fistulous connection between the circumflex artery and the left bronchial artery was noted (Figure 3A, 3B, 3C, and 3D). CT-coronary angiogram as further done to delineate the fistulous tract, which showed a long, tortuous, fistulous connection between left circumflex artery and left bronchial artery. Distal segment of the fistula gives multiple dilated tortuous branches just before joining the left bronchial artery (Figure 4A, 4B, and 4C). High resolution plain CT of bilateral lungs showed normal parenchyma (Figure 5). As the symptoms were mild, we opted for optimal medical management for the patient with diltiazem, oral nitrate, and followed her up for worsening of symptoms. At a 6-month follow-up the patient was completely asymptomatic.

Discussion

A coronary-artery fistula (CAF) is a rare anomalous communication between a coronary artery and a cardiac chamber or major vessel.[1] CAF accounted for 13% of coronary artery anomalies.[6] CAF is detected in 0.18% of coronary angiographies. The most common fistula being coronary artery–pulmonary artery fistula, which is often

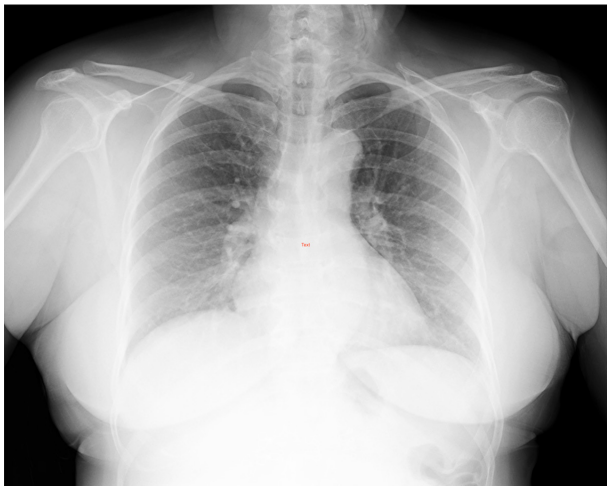

Figure 1

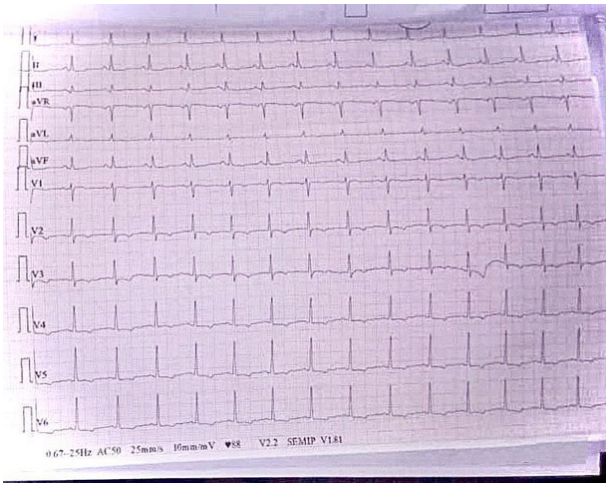

Figure 2

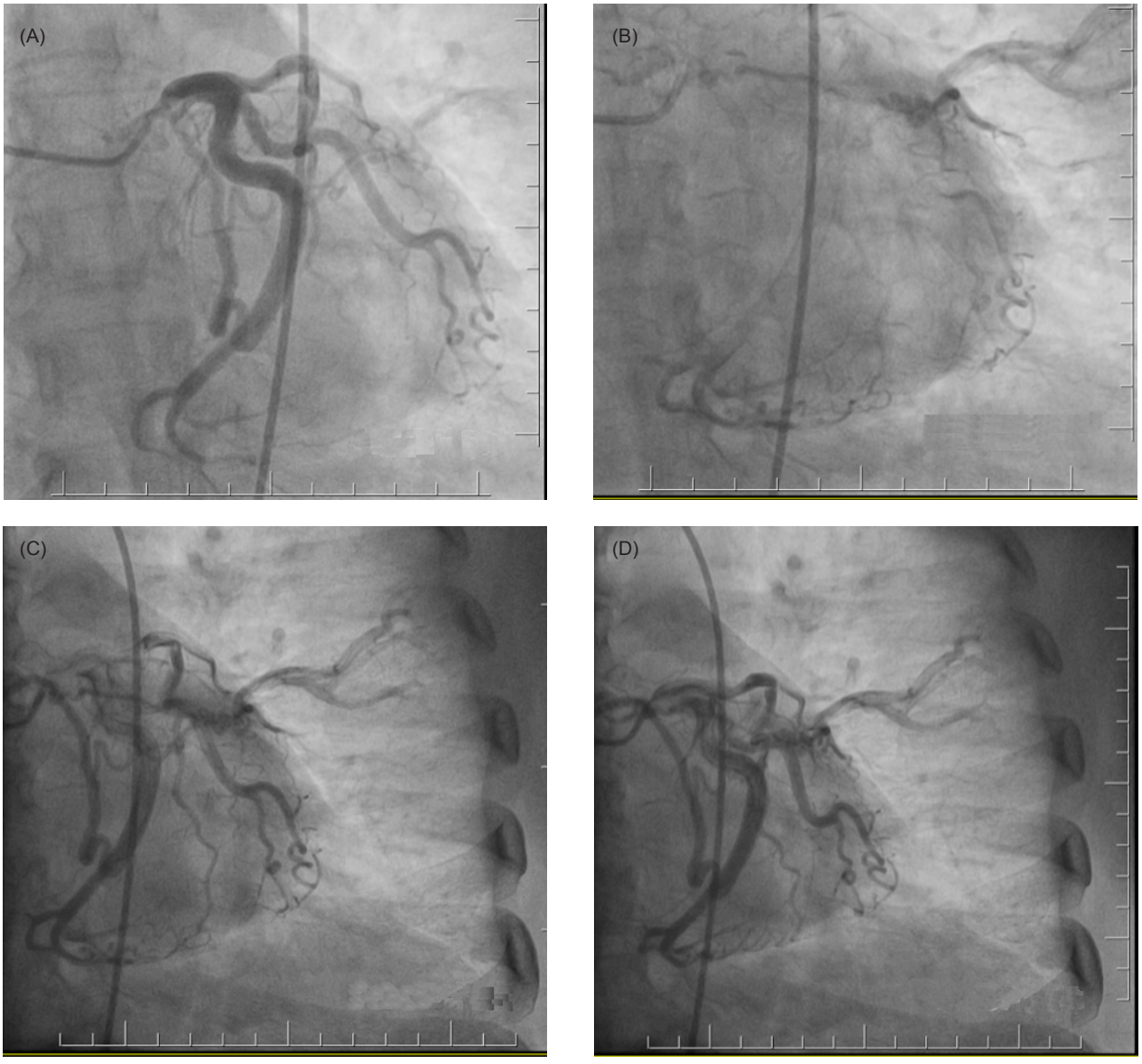

Figure 3

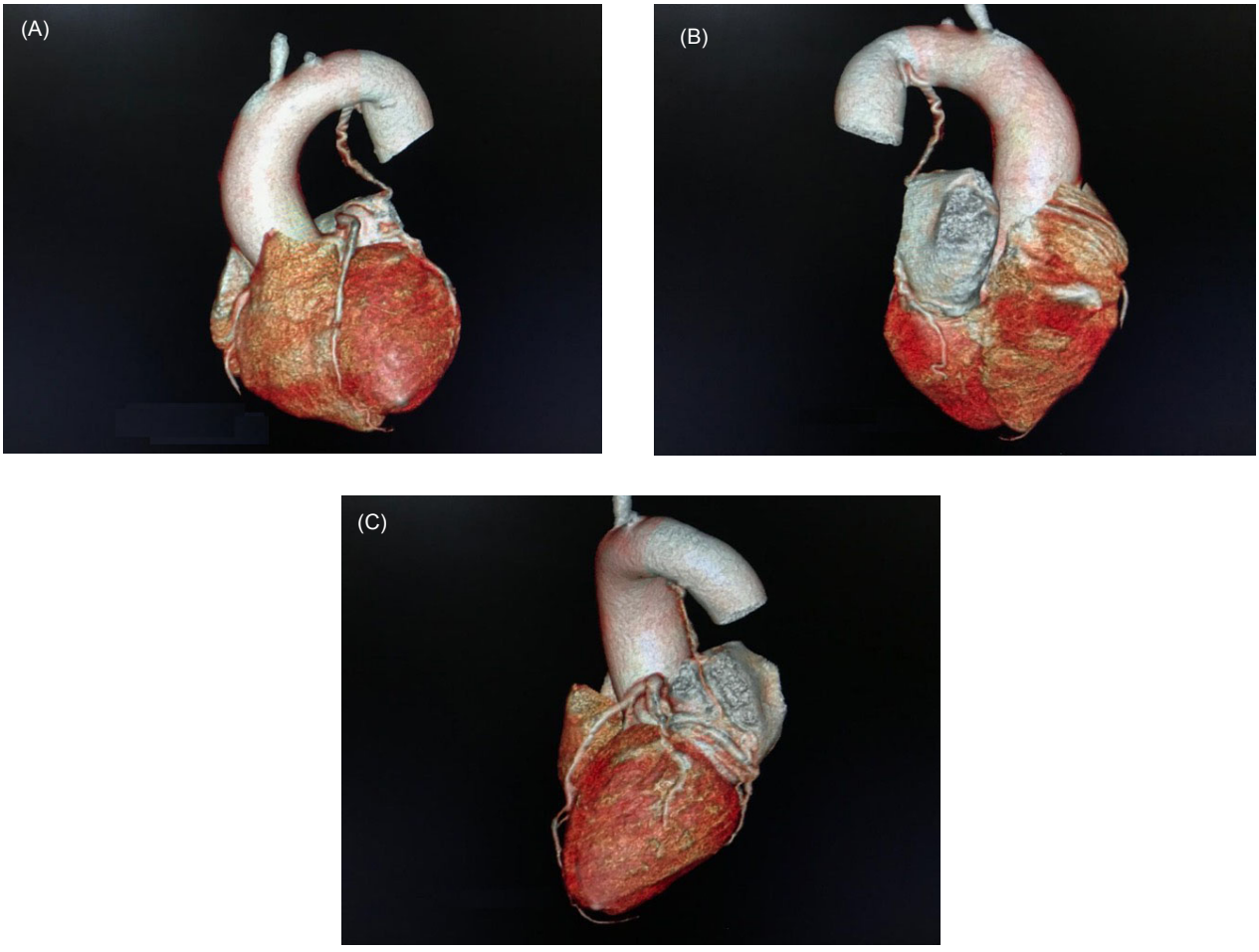

Figure 4

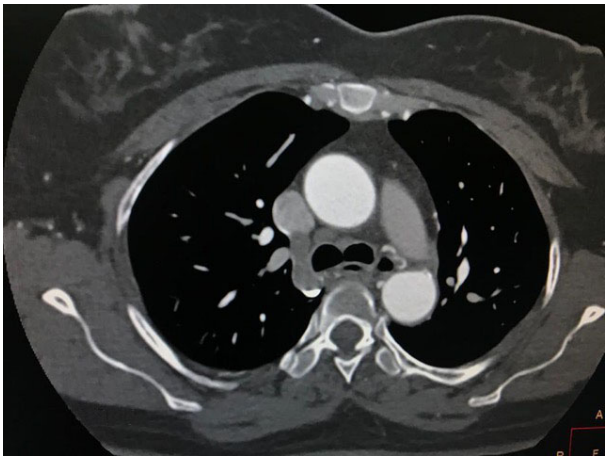

Figure 5

associated with bronchiectasis. Majority of them are congenital, but rare cases of traumatic or iatrogenic origin: after coronary intervention or valve replacement are reported.[3] Other bronchopulmonary disease like chronic obstructive airways disease, multiple bullae, and cystic formations are seldom associated with CAF.[2] Coronary-bronchial artery fistula (CBAF) is extremely rare. It is observed that CBAFs are present since birth, but they majority remain closed since filling pressure of coronary and bronchial circulations remain almost the same. Several mechanisms are postulated to be involved in pathogenesis of CBAF: various cardiovascular diseases, Takayasu arteritis, pulmonary thromboembolism, bronchiectasis, and pulmonary tuberculosis.[5]

More than 90% of CAFs open into right heart chambers or their connecting vessels. About 40% connect to the right ventricle, 25% to right atrium, 15% to 20% to pulmonary artery, 7% to coronary sinus, and only 1% to superior vena cava[7]. Fistulas to the left ventricle are very rare, with an incidence of only 3%.[8] CBAFs typically arise from left circumflex artery as seen in this case. In a review article by Said et al.,[9] the origin of the CBAFs was reported to be the left circumflex artery in 61%, the right coronary artery in 36%, and the left anterior descending artery in 3%.

Clinical features are varied and depend on the type of fistula, shunt volume, site of the shunt, and presence of concomitant cardiac/pulmonary abnormalities. Majority remain asymptomatic. Angina and/or dyspnea on exertion are the most common symptoms and the underlying mechanism is likely coronary steal phenomenon[2] or very rarely, rupture of an aneurysmal segment. Hemoptysis is an extremely rare and life threatening symptom. As the degree of the left-to-right shunt increases, complications such as pulmonary hypertension, infective endocarditis, or congestive heart failure occur more frequently. A few cases of rupture or thrombosis of coronary artery fistulas have been reported in association with arterial aneurysms or coronary steal phenomena.[6]

Echocardiography is essential for the evaluation of concurrent cardiac anomalies and left-to-right shunts. Diagnosis is mainly established by invasive coronary angiography. Contrast enhanced CT has high diagnostic accuracy and can reveal the extent and drainage of fistulas adequately. In this case, contrast CT is of value in detecting the course and confirming CBAF with high accuracy. The functional assessment may be obtained by myocardial perfusion imaging (MPI) using technetium-99 m tetrofosmin, MRI, and oximetric series during cardiac catheterization to establish the magnitude of the shunt.

Although management of CBAF is still debated, optimal medical therapy is generally considered the first line of treatment for asymptomatic or minimally symptomatic cases. Most data are obtained mainly from anecdotal case reports or small series of cases. Surgical or percutaneous correction may be considered only in symptomatic patients with heart failure, myocardial ischemia, or high-flow shunting or those patients who are unresponsive to medical therapy. Presence of concurrent cardiac anomalies should also be taken into consideration. Shin et al.[8] proposed prophylactic and therapeutic embolization of CBAF in patients with bronchiectasis. Both surgical and percutaneous corrections have a high incidence of complication and failure rate, especially when the fistulas are associated with aneurysmal dilatation or multiple branching patterns. Coil embolization is preferred when there is evidence of significant coronary steal phenomenon. Myocardial infarction is a dreaded post procedural complication.

Conclusion

CBAF is an extremely rare cause of typical angina per se. Multidetector computed tomography (MDCT) is a useful supplementary investigation for anatomical orientation of CBAF especially where interventions are planned. Optimal medical management is a viable option in cases where the symptoms are mild, limiting the use of potentially unsafe invasive procedures.

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