Rare congenital cardiac anomaly presenting with predominant respiratory complaint: A case report

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ABSTRACT

Introduction: Coronary cameral fistula is a rare anomaly where there is a fistulous connection between coronary artery and one of the cardiac chambers. It generally presents with predominant cardiac complaints or as accidental finding in an asymptomatic individual. Our patient presented chiefly with respiratory complaint and was misdiagnosed as tuberculosis.

Case Report: A 22-year-old female presented in chest OPD with complains of cough and fever since one month and was started on anti-tubercular treatment since one month. On examination there was continuous murmur, thrill and coarse crackles in right axillary area. On Chest X-ray there was a multi-lobulated mass in the right side with upper zone linear and cystic opacities. Computed tomography (CT) scan of Thorax showed right middle lobe bronchiectasis and a large fistula arising from right coronary artery and draining into right superior pulmonary vein and left atrium. Cardiac catheterization confirmed the diagnosis and coronary arteriovenous fistula closure was done by percutaneous route via lifetechcera vascular plug.

Conclusion: Coronary cameral fistula despite being large in size may remain silent without causing any hemodynamic abnormality or any compromise in the cardiac function. The predominant complaint may be due to pressure effect of the aneurysmal vessel on adjacent bronchi and causing secondary changes in the pulmonary parenchyma fed by these bronchi. A thorough clinical examination may clinch the diagnosis.
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How to cite this article

Chaurasia S, Kumar R, Haran A, Katare S. Rare congenital cardiac anomaly presenting with predominant respiratory complaint: A case report. Int J Case Rep Images 2015;6(11):678–681.
doi:10.5348/ijcri-2015107-CR-10568

INTRODUCTION

A sizable communication between one of the branches of coronary artery and a chamber of heart bypassing the myocardial capillary bed is called coronary cameral fistula. It is a rare congenital anomaly with incidence of 0.08–0.3% of patient undergoing routine diagnostic left heart catheterization [1, 2]. Most of the small fistulae
are clinically silent [1]. Large fistulas progressively enlarge over time and are more likely to cause cardiac complication like congestive heart failure, myocardial infarction, arrhythmias, aneurysm formation, rupture and death [1, 3, 4]. Majority of the symptomatic patients present with symptoms like exertional dyspnea, angina, fatigue, palpitations and syncope often pointing to the cardiac origin of symptoms.

Most of the cases ECG and chest X-rays are not helpful in diagnosis and Echocardiography and cardiac catheterization and angiography confirm the diagnosis. Chest X-ray findings are generally normal except in the presence of significant shunt flow when cardiomegaly may be evident [5]. In addition pulmonary venous congestion and interstitial edema may be seen. ECG may reveal effect of volume overload in presence of coronary steal, ischemic changes and/or arrhythmias. Herein, we report a case of coronary cameral fistula presenting chiefly with respiratory complaints and chest X-ray abnormality and was wrongly diagnosed as tuberculosis.

CASE REPORT

A 22-year-old female presented in chest outpatient department with chief complaints of cough with expectoration, intermittent fever and breathlessness on exertion since three months. Patient also gave history of palpitation and easy fatigability. Patient was multipara with two uneventful pregnancies and both kids were delivered at full term by normal delivery. She was started on anti-tubercular treatment since one month by general practitioner on the basis of chest X-ray abnormality. Patient had history of recurrent cough with expectoration since last two years which responded to antibiotics prescribed by general practitioner.

Physical examination was remarkable with presence of grade 4 continuous murmur over right mammary, axillary and infra axillary areas, moreover breath sounds were decreased in right lower inter scapular area and infra scapular area and coarse mid and late inspiratory crackles could be appreciated in the right axillary region.

On investigation chest X-ray showed multi lobulated opacities arising from the right hilar region and extending into right middle and lower zones. There was presence of linear and cystic opacities in right upper zone (Figure 1 and 2). ECG was normal. 2D echocardiography showed dilated right coronary artery draining into left atrium with dilated left ventricle, mild mitral regurgitation and mild aortic regurgitation. Cardiac multidetector computed tomography scan showed grossly dilated and tortuous right coronary artery having fistulous communication with the dilated venous sac which in turn drains into superior pulmonary vein and left atrium. One of the largest sac was measuring 7x5 cm in size (Figure 3). Venous sacs were seen compressing right middle lobe bronchus resulting in bronchiectasis of right middle lobe (Figure 4).

Sputum was negative for acid-fast bacillus and Gram stain and culture sensitivity of sputum yielded Pseudomonas aeruginosa. Anti-tubercular treatment was stopped and was started on appropriate antibiotics, mucolytic agent and chest physiotherapy. Patient responded to treatment and was asymptomatic after 10 days of antibiotics. Follow-up culture was negative. Patient was referred to cardiologist for diagnostic cardiac catheterization which showed large coronary fistula.
arising from right coronary sinus to left atrium through a long tortuous tunnel passing through superior pulmonary vein.

Coronary arteriovenous fistula closure was done by percutaneous route via lifetechcera vascular plug 16 mm, once the infection was controlled. Post deployment check angiogram showed mild residual shunt. Patient is asymptomatic after undergoing fistula closure.

**DISCUSSION**

A coronary cameral fistula is a rare vascular anomaly found in approximately 0.08–0.3% of patients undergoing diagnostic coronary angiography [1, 2]. Ninety percent of coronary cameral fistulas drain into right sided chambers of the heart [6]. Clinical presentation generally depend on the hemodynamic significance of the anomaly with majority of small fistulas found incidentally in asymptomatic individuals [1]. Patients with large shunts often present with symptoms arising due to myocardial steal causing angina, dyspnea, congestive heart failure and arrhythmias [1, 3, 4]. The cornerstone in establishing the diagnosis is 2D echocardiography and diagnostic cardiac catheterization and coronary angiography. In majority of cases, chest X-ray is normal or cardiomegaly is there, abnormal chest X-ray shadow is seen in approximately 4% of cases [5]. Our case is unique in many ways, firstly the predominant complaints of the patient was cough with expectoration and fever and chest X-ray was showing abnormal shadows in the right mid and lower zone without any obvious cardiomegaly, in view of above the patient was started on AKT by private practitioner. Secondly, the fistula was draining into the left side of the heart and despite there being huge aneurysmal dilatation of the right coronary artery there was no hemodynamic disturbance and the only complication was arising due to pressure effect of the aneurysmal sac over right middle lobe bronchus causing bronchiectasis. In our case physical examination gave a clue to the diagnosis which was later confirmed by more sophisticated investigations.

**CONCLUSION**

All abnormal shadows on chest X-ray in a patient with respiratory complaint need not be originating from the lung. A thorough clinical examination and investigations should be done before labeling abnormal shadows as tuberculosis. Coronary cameral fistula despite being large in size may remain silent without causing any hemodynamic abnormality or any compromise in the cardiac function. The predominant complaint may be due to pressure effect of the aneurysmal vessel on adjacent bronchi and causing secondary changes in the pulmonary parenchyma fed by these bronchi. A thorough clinical examination may clinch the diagnosis.
Guarantor
The corresponding author is the guarantor of submission.

Conflict of Interest
Authors declare no conflict of interest.

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