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

Sheetal Chaurasia, Ravi Kumar, Alamelu Haran, Srikanth Katare

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

International Journal of Case Reports and Images (IJCRI)

International Journal of Case Reports and Images (IJCRI) is an international, peer reviewed, monthly, open access, online journal, publishing high-quality, articles in all areas of basic medical sciences and clinical specialties.

Aim of IJCRI is to encourage the publication of new information by providing a platform for reporting of unique, unusual and rare cases which enhance understanding of disease process, its diagnosis, management and clinico-pathologic correlations.

IJCRI publishes Review Articles, Case Series, Case Reports, Case in Images, Clinical Images and Letters to Editor.

Website: www.ijcasereportsandimages.com

CASE REPORT

PEER REVIEWED | OPEN ACCESS

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

Sheetal Chaurasia, Ravi Kumar, Alamelu Haran, Srikanth Katare

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.

Keywords: Arteriovenous fistula, Congenital anomaly, Coronary cameral fistula, Middle lobe bronchiectasis, Respiratory involvement

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

Sheetal Chaurasia¹, Ravi Kumar², Alamelu Haran³, Srikanth Katare⁴

Affiliations: ¹Assistant Professor, Department of respiratory medicine, Vydehi Institute of Medical Sciences and Research Centre, Bangalore, Karnataka, India; ²Associate Professor, Department of Respiratory Medicine, Vydehi Institute of Medical Sciences and Research Centre, Bangalore, Karnataka, India; ³Professor and Head, Department of Respiratory Medicine, Vydehi Institute of Medical Sciences and Research Centre, Bangalore, Karnataka, India; ⁴Post graduate, Department of respiratory medicine, Vydehi Institute of Medical Sciences and Research Centre, Bangalore, Karnataka, India.

Corresponding Author: Sheetal Chaurasia, Department of Pulmonary Medicine, Vydehi Institute of Medical Sciences # 82, EPIP Area, Whitefield, Bangalore, Karnataka, India - 560066; Ph: +919972949900; +9108028412956; E-mail: sheetal.utkalit@gmail.com

Received: 05 June 2015

Accepted: 01 July 2015

Published: 01 November 2015

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).

Figure 1: Chest X-Ray showing multi-lobulated opacity in right mid and lower zone. Cystic changes in right upper zone can be appreciated.

Figure 2: CT Scout Film - showing multi-lobulated mass in right lung.

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

Figure 3: Contrast enhanced cardiac computed tomography In multidetector computed tomography scanner- grossly dilated right coronary artery having fistulous communication with dilated venous sac draining into left atrium. Largest sac measures 7x5 cm. wall calcification can be appreciated suggesting long standing process.

Figure 4: Venous sacs are compressing the right middle lobe bronchus resulting in bronchiectasis of the right middle lobe.

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 accidentally 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.

Author Contributions

Sheetal Chaurasia – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Ravi Kumar – Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Alamelu Haran – Substantial contributions to conception and design, Revising it critically for important intellectual content, Final approval of the version to be published

Srikanth Katare – Acquisition of the data, Drafting the article, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

Copyright

© 2015 Sheetal Chaurasia et al. This article is distributed under the terms of Creative Commons Attribution License which permits unrestricted use, distribution and reproduction in any medium provided the original author(s) and original publisher are properly credited. Please see the copyright policy on the journal website for more information.

2. Yamanaka O, Hobbs RE. Coronary artery anomalies in 126,595 patients undergoing coronary arteriography. *Cathet Cardiovasc Diagn* 1990 Sep;21(1):28–40.
3. Brooks CH, Bates PD. Coronary artery-left ventricular fistula with angina pectoris. *Am Heart J* 1983 Aug;106(2):404–6.
4. McLellan BA, Pelikan PC. Myocardial infarction due to multiple coronary-ventricular fistulas. *Cathet Cardiovasc Diagn* 1989 Apr;16(4):247–9.
5. Said SA. Current characteristics of congenital coronary artery fistulas in adults: A decade of global experience. *World J Cardiol* 2011 Aug 26;3(8):267–77.
6. Stierle U, Giannitsis E, Sheikhzadeh A, Potratz J. Myocardial ischemia in generalized coronary artery-left ventricular microfistulae. *Int J Cardiol* 1998 Jan 5;63(1):47–52.

REFERENCES

1. Vavuranakis M, Bush CA, Boudoulas H. Coronary artery fistulas in adults: incidence, angiographic characteristics, natural history. *Cathet Cardiovasc Diagn* 1995 Jun;35(2):116–20.

Access full text article on
other devices

Access PDF of article on
other devices

Edorium Journals: An introduction

Edorium Journals Team

About Edorium Journals

Edorium Journals is a publisher of high-quality, open access, international scholarly journals covering subjects in basic sciences and clinical specialties and subspecialties.

Invitation for article submission

We sincerely invite you to submit your valuable research for publication to Edorium Journals.

But why should you publish with Edorium Journals?

In less than 10 words - we give you what no one does.

Vision of being the best

We have the vision of making our journals the best and the most authoritative journals in their respective specialties. We are working towards this goal every day of every week of every month of every year.

Exceptional services

We care for you, your work and your time. Our efficient, personalized and courteous services are a testimony to this.

Editorial Review

All manuscripts submitted to Edorium Journals undergo pre-processing review, first editorial review, peer review, second editorial review and finally third editorial review.

Peer Review

All manuscripts submitted to Edorium Journals undergo anonymous, double-blind, external peer review.

Early View version

Early View version of your manuscript will be published in the journal within 72 hours of final acceptance.

Manuscript status

From submission to publication of your article you will get regular updates (minimum six times) about status of your manuscripts directly in your email.

Our Commitment

Six weeks

You will get first decision on your manuscript within six weeks (42 days) of submission. If we fail to honor this by even one day, we will publish your manuscript free of charge.

Four weeks

After we receive page proofs, your manuscript will be published in the journal within four weeks (31 days). If we fail to honor this by even one day, we will publish your manuscript free of charge and refund you the full article publication charges you paid for your manuscript.

Most Favored Author program

Join this program and publish any number of articles free of charge for one to five years.

Favored Author program

One email is all it takes to become our favored author. You will not only get fee waivers but also get information and insights about scholarly publishing.

Institutional Membership program

Join our Institutional Memberships program and help scholars from your institute make their research accessible to all and save thousands of dollars in fees make their research accessible to all.

Our presence

We have some of the best designed publication formats. Our websites are very user friendly and enable you to do your work very easily with no hassle.

Something more...

We request you to have a look at our website to know more about us and our services.

We welcome you to interact with us, share with us, join us and of course publish with us.

Edorium Journals: On Web

Browse Journals

CONNECT WITH US