Case Report

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Submitral Aneurysm with Tuberculous Endocarditis of Mitral Valve: An Extremely Uncommon Presentation of A Common Disease

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Abstract

Submitral aneurysm is an extremely rare cardiac anomaly which was initially described among the African population and is considered rare in other parts of the world. In majority of the reported cases aetiology is congenital. We report a unique case of 54-year-old immunocompetent female with submitral aneurysm and tuberculous endocarditis of the mitral valve. She underwent successful submitral aneurysmoraphy with mitral valve replacement. Postoperative recovery has been uneventful without relapse for 12 months.

Keywords: Submitral aneurysm; Severe mitral regurgitation; Tuberculous endocarditis.

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Introduction

Submitral LV aneurysm is an extremely rare cardiac anomaly which was first described by Corvisart in the year 1812.[1] This condition has been predominantly described in African population of the world, extremely rare in individuals of non- African ethnicity.[2] Although majority of the cases are congenital few cases due to Takayasu arteritis and Tuberculosis have been reported.[3,4] In 1826, Laennec was the first to describe cardiac tuberculosis assigning heart as the 13th organ affected in order of frequency.[5] In a large series of 7683 cases tuberculous myocardial involvement was present only in 0.64% of cases.[6] Tuberculous endocarditis usually manifests in the context of immunodeficient persons and miliary tuberculosis.[7]

Unique features of our case include disease in immunocompetent patient, without evidence of pulmonary or miliary tuberculosis, non–African race, histological evidence of tuberculosis and successful surgical outcome without recurrence after completion of ATT. To the best of our knowledge only 3 cases of tuberculous subvalvar aneurysms have been reported before in immuno competent individuals without miliary TB.[8–10]

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Case Report

A 54 year female, diagnosed as a case of severe mitralregurgitation was admitted with us for mitral valve replacement. She had complaints of breathlessness on excertion NYHA Class II, since 3 months. No history of fever or RHD. Clinical examination revealed a regular pulse of 86/min, blood pressure 130/70 mm Hg. Her electrocardiogram showed sinus rhythm, features of LA enlargement and LV volume overload. Her laboratory investigations demonstrated a normal hemogram except for raised ESR, liver and renal function tests were within normal limits. Her chest radiograph revealed mild cardiomegaly. Transthoracic echocardiography revealed the exact cause of sub-mitral aneurysm severe mitral regurgitation as a large measuring 7 × 5 × 4 cms, filled with clots arising immediately below the posterior mitral leaflet. Additionally TEE revealed grossly thickened AML with cystic lesion in it. Color Doppler flow imaging demonstrated systolic flow into the aneurysm and mitral regurgitation. She had mild tricuspid regurgitation as well with normal systolic left ventricular function. Her coronary angiogram demonstrated normal coronaries. The cardiac CT revealed submitral aneurysm measuring 7 × 5.5 × 4.2 cm in relation to lateral wall of LV. Neck of the aneurysm measured 3 cm and aneurysm was filled with clots. LCX was displaced anterosuperiorly without luminal compression. Diagnosis of a submitral aneurysm causing severe mitral regurgitation was established with suspected thickening of AML pathology of it was unknown preoperatively. The patient was scheduled for elective surgical repair of the aneurysm with mitral valve replacement due to suspicious lesion in the AML.

Intraoperatively complete invasive and non-invasive monitoring including transesophageal echocardiography was established. After a midline sternotomy and pericardiotomy, the anatomy of the aneurysm was identified. It was arising from the postero basal wall of the left ventricle with a broad neck **Fig. 1**. After systemic heparinization, cardiopulmonary bypass was established using aortic and bicaval cannulation. Under cardioplegic arrest and moderate hypothermia, LA was opened parallel to waterson's groove to note, grossly thickened AML, retracted PML, mouth of the aneurysm underlying the entire length of posterior mitral annulus with intervening ridge dividing the mouth into two **Fig. 2**. Aneurysm was filled with clots.

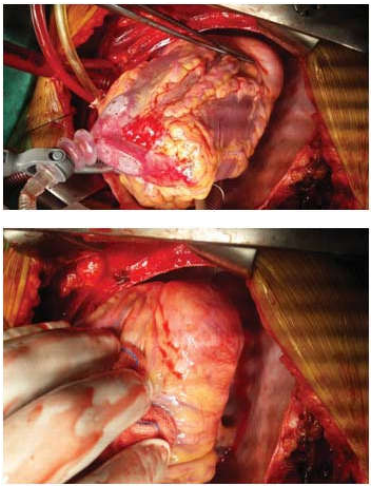

Fig 1: Intraoperative view of submitral aneurysm–externally.

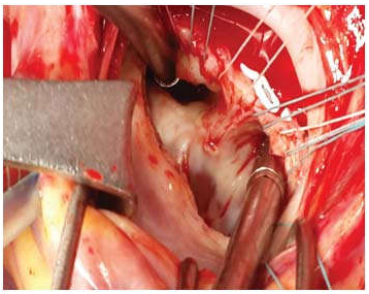

Fig 2: Showing the orifice of submitral aneurysm after excision of mitral leafelets

All clots were removed, AML and PML were excised. Mouth of the aneurysm was closed with PTFE patch using 2–0 pledgetted TYCRON sutures **Fig. 3**. MVR was done with 27 mm St. Jude mechanical valve **Fig. 4**. LA was closed in two layers. Later aneurysm was opened externally to confirm the completeness of the patch and residual sac was excised and aneurysmoraphy was done. After adequate rewarming, the patient was successfully weaned off cardiopulmonary bypass. Transesophageal echocardiography demonstrated successful closure of the communication between left ventricle and aneurysmal sac. The normal functioning of the prosthetic mitral valve were conclusive of complete surgical correction.

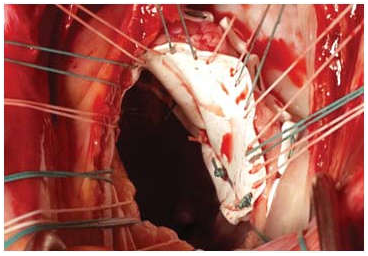

Fig 3: Patch closure of the submitral aneurysm.

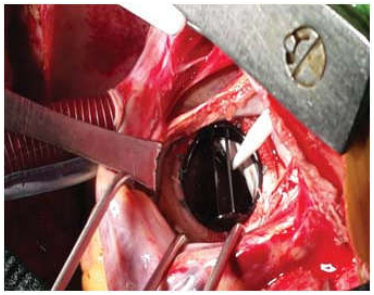

Fig 4: Post-mplantation of mitral valve- PTFE patch can be seen through the valve.

Postoperatively patient recovered normally and was uneventful. Interestingly biopsy of the leaflets revealed epitheloid granulomas with caseating necrosis pointing towards tuberculous aetiology. Subsequently patients was evaluated and had no foci of pulmonary or military tuberculosis. In concurrence with a pulmonologist patient was started on ATT and she has completed the course successfully without recurrence. She is under regular followup for the past 12 months without relapse. She has been anticoagulated with Warfarin for her lifetime with an objective of maintaining INR between 2.5–3.5 in view of her mechanical valve prosthesis.

Discussion

Submitral Aneurysm is a rare lesion that occurs most often in the black population. The prevalence of this lesion among blacks[11–13] appears to indicate a congenital origin or predisposition.[12] Congenital Submitral aneurysm seems to be caused by a junctional defect between the cardiac muscle and the fibrous structure of the heart.[13] They have also been described as a complication in patients with infective endocarditis, tuberculosis and syphilis.[14–17] Most of the few reported cases due to tuberculosis have occurred in patients who were immunodeficient or had military tuberculosis.[7] Unique features of our case include disease in immunocompetent patient, without evidence of pulmonary or miliary tuberculosis.

The anatomy of the submitral aneurysm is quite complex and its size can vary from a few millimeters up to several centimeters. It can extend behind the left atrium, the left ventricle or both.[13] Pathophysiologically, sub-mitral aneurysm involves the fibrous mitral annulus and with enlargement displaces the posterior mitral annulus and sub-valvular supporting apparatus apart, resulting in restriction of the posterior mitral leaflet and failure of leaflet coaptation with secondary mitral regurgitation.[13] Finally, loss of support for the posterior leaflet, together with cardiac failure due to systolic expansion of the aneurysm, worsens mitral regurgitation.

Clinical symptoms arise as a result of valvular regurgitation or occasionally from compression of cardiac structures.[13] Because a submitral aneurysm can be asymptomatic for many years; it may not be detected unless routine echocardiography is performed.[18] In our patient presentation was in the form of shortness of breath on exertion probably due to the development of mitral regurgitation. Sometimes, the first sign of the disease is ventricular tachycardia, central or peripheral embolism or sudden death[13].

Diagnosis by chest X-ray is easy if the calcification is present in the aneurysm wall.[19] At present, the transthoracic and transesophageal echocardiography are the most accurate diagnostic tools.[18] The typical location of the aneurysm and the absence of coronary artery disease on angiography confirms the diagnosis of submitral aneurysm. Surgical treatment is mandatory as soon as the diagnosis of submitral aneurysm is made, inorder to treat the cardiac failure that arises from expansion of the aneurysm and to avoid other potential cardiovascular events. Surgical intervention is the only method of treatment for submitral aneurysm. The surgical approach can be extracardiac, intracardiac or combined. The first surgical correction was reported in 1963 by Shrire and Bernard through extracardiac approach.[20,21] A series of 9 patients who underwent surgical repair of submitral aneurysm was reported by Antunes in 1987 by intracardiac approach.[13] Surgical repair with intracardiac approach and combined approach was reported in a series of 12 patients by Du Toit HJ *et al.*[16] An innovative technique in which the aneurysm was approached via left atrium with detachment of posterior mitral leaflet has been recently described.[22] Successful surgical repair is dependent on the appropriate understanding of the anatomy of the aneurysm, its relation with mitral valve and annulus.

In our patient, we have successfully used the combined approach for surgical correction of submitral aneurysm. Combined approach is the preferred technique in submitral aneurysms because of the extent of posterior mitral annulus involvement, size of the aneurysm and degree of mitral regurgitation.

While addressing the issue of mitral regurgitation, if it is due to annular dilatation as seen in majority

of the cases MV repair can be attempted, but if the leafelets are thickened as in our case and infective aetiology is suspected it is better to do valve replacement.

Conclusion

Submitral aneurysms are extremely rare, but tuberculosis should be considered as one of the aetiological factors in Indian population, histological and microbiological evidence should be sort to confirm the diagnosis. Initiation of ATT compliments good surgery in avoiding recurrence.

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