

Title of MMed Dissertation:

**Left Ventricular Submitral
Aneurysms**

Candidate

Dr Henning J Du Toit

Student Number: DTTHEN005

**Chris Barnard Division of Cardiothoracic Surgery
University of Cape Town**

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Promotors/Supervisors

Dr John Hewitson

Dr John Lawrenson

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Objective: Retrospective institutional review of the pathology, aetiology, classification and surgical management of left ventricular submitral aneurysms (LVSMA). These aneurysms are a well recognized but relatively rare disease found commonly in patients from African ancestry.

Methods: The series comprises 20 consecutive patients treated surgically at three institutions from 1985 to 2002. Natural history, clinical presentation, histo-pathological findings, suspected aetiology, operative techniques, along with a discussion of the condition is presented.

Results: There were 10 female and 10 male patients and the mean age was 17 ± 6 (range 8 – 34) years. Patients were grouped as to the degree of posterior mitral annulus involvement by the aneurysm. In Group I, (n = 12) a single aneurysm neck was found. In Group II, (n = 3) multiple necks and in Group III, (n = 5) involvement of the entire posterior annulus by the aneurysm was found. Mean age in Group III (29 ± 5 years) was older than that of Groups I and II (15.5 ± 4 years) suggesting a progressive nature of these aneurysms to enlarge. Clinically patients were in New York Heart Association (NYHA) class I – IV. An intra-cardiac surgical approach was used in 11, extra-cardiac approach in two and a combined approach in seven patients. Mitral valve repair was attempted in 14 patients, with two intra-operative mitral valve repair failures. Failure to control the aneurysm neck (n = 2) and failure of mitral valve repair (n = 2) resulted in subsequent re-operation. There was no operative mortality. Histology of the aneurysm tissue suggested co-existing rheumatic heart disease in two, tuberculosis in four and infective endocarditis in two. Unknown or congenital disease was postulated in nine patients. Although LVSMA are thought to be congenital, 8 out of 20 patients (40%) had evidence of co-existent inflammatory pathology.

Conclusion: The etiology of LVSMA remains uncertain. Many are thought to be congenital, but the findings in this study strongly support the view that rheumatic disease, chronic infections and malnutrition also play a role. A new classification is proposed based on the pathological findings. Involvement of the entire annulus in the older patients suggests a possible progressive nature of the disease. Surgery should be the definitive therapy in all patients. Surgical approach must be individualized but the intra cardiac approach is suitable for the surgical repair in most

cases. Success in management is dependent on the appropriate understanding of the relationship between aneurysm and valve.

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INTRODUCTION

Background

The entity of non-ischaemic, congenital left ventricular submitral aneurysm (LVSMA) is well recognized, but is not mentioned in most standard textbooks. These aneurysms are found immediately below the posterior leaflet of the mitral valve and have necks of varying size. They occur almost exclusively in patients from black African ethnic origin, particularly in Africa south of the Sahara [1,2,3,4]. They have been documented by researchers mostly in Nigerians and the Southern Africans, and instances among North American Blacks and West Indians have been reported [1 - 8]. LVSMA are occasionally seen in Caucasians and people of mixed race, but to a lesser degree [3,9]. Other terms used in the literature to describe these aneurysms are: sub-annular, subvalvar, submitral valvular, non-ischaemic sub-annular, "Bantu" aneurysm and supernumerary chambers of the left ventricle.

The eminent British Surgeon and Physician, John Hunter, was the first to describe Left Ventricular Aneurysms (LVA) but the distinction between subvalvar aneurysms and LV free wall aneurysms was by all accounts not made [1]. Submitral aneurysms were first described by Corvisart in 1812 [2]. Reports in recent times came mainly from research done by American and African researchers in the late 1950's and early 1960's. [1,2,3,5,6]. Reports by Schlicter, Chesler and Wolpowitz in the 50's, 60's and 70's [2,3,8] provide comprehensive descriptions of the major clinical features of this disorder. An institutional review was published by Du Toit in 2005 discussing recent advances in diagnostics, classification and surgical treatment [10]. Building on the above data this thesis is presented. A number of etiologies were proposed for the development of the anatomical and patho-physiological processes associated with the disease and it is thought that these aneurysms are due to a congenital weakness in the annulus [1-5,9,10]. Patho-physiologically, sub-mitral aneurysms involve the fibrous mitral annulus, and with enlargement, they displace the posterior mitral leaflet, annulus and subvalvar supporting apparatus. This results in restriction of the posterior mitral leaflet and failure of leaflet

co-aptation with secondary mitral regurgitation [2,9,1*]. Clinical⁷ symptoms arise as a result of valvar regurgitation or occasionally from compression of cardiac structures [2,3,6,8,14,1*].

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Object of Study

The object of this study was to consolidate and evaluate all the information available on the disease. A number of new conclusions are presented based on the clinical evidence found in the patients studied. The study reviews the collective experience from three institutions, namely, Red Cross War Memorial Children Hospital, Groote Schuur Hospital in Cape Town and Wentworth Hospital in Durban. Diagnosis and surgical approach, a proposed classification, and complications experienced with the repair of sub-mitral aneurysms will be discussed in detail.

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STUDY

1. Methods

1.1 Patients

A retrospective review was undertaken of 20 consecutive patients referred to Groote Schuur, Red Cross Children and Wentworth Hospitals, for surgical correction of LVSMA between 1985 and 2002. The possible aetiology of LVSMA, clinical presentation, laboratory results, surgical findings and histopathology were examined. Medical history and clinical presentation based on Tuberculosis Score Chart A (International Union Against Tuberculosis and Lung Disease) was used to diagnose tuberculosis in the patients from Wentworth Hospital (n=8)[11]. In patients from the other two institutions the clinical presentation, together with positive microbiology, was used to diagnose tuberculosis (n=12). Tissue samples of all the patients that were thought to have tuberculosis were sent for histology to confirm the etiology. In patients with clinical signs of infective endocarditis, blood cultures were used to diagnose and confirm infective endocarditis. In patients with clinical signs of rheumatic fever, serology (antistreptolysin O titre (ASOT) and histology was used to diagnose and confirm Rheumatic Fever.

On macroscopic inspection of the annulus during surgery patients were divided into three groups according to the extent of posterior mitral annulus involvement by the aneurysm neck:

Type I, single localized neck;

Type II, multiple necks (separate distinct openings, and not a large neck with pseudo division by the sub-mitral chordal apparatus into separate openings); Type III, involvement of the entire posterior mitral annulus.

1.2 Surgery

Cardiopulmonary bypass with bicaval venous cannulation and moderate hypothermia (30 -32°C) was used in all cases. Myocardial protection, after aortic cross clamping, was provided with cold St Thomas No. 2 crystalloid cardioplegic solution with added lignocaine (20mg/l) for initial arrest and intermittent cold 1:1 St Thomas/blood cardioplegia re-infusions for maintenance. Two techniques to approach and repair sub-mitral aneurysms were used. In the intra-cardiac approach, the left atrium and mitral valve was approached via an incision through the inter-atrial groove. In the extra-cardiac approach, an epicardial incision is used to open the aneurysm.

Using the intra-cardiac approach, an incision into the left atrium through the inter-atrial groove was made. The neck of the sub-mitral aneurysm was identified by retracting the edge of the posterior mitral leaflet, and examining the sub-valvar area between the mitral valve annulus and papillary muscles. A 'right-angled' instrument was then inserted into the aneurysm to determine the relationship between the aneurysm and the posterior left atrial wall. An incision was made in the floor of the left atrium parallel to the posterior mitral annulus to allow access to the aneurysm neck. Alternatively the posterior mitral leaflet can be detached from the posterior mitral annulus, to visualize the aneurysm neck. The mitral leaflets are usually normal and the area and height of the posterior mitral leaflet needs to be preserved. The advantage of this 'intracardiac' approach is that the mitral valve can be assessed and tested for competence following repair of the mitral valve.

In the extra-cardiac approach, the heart was displaced by retracting it anti-clockwise to expose the aneurysm. The aneurysm is found in the vicinity of the left atrial appendage posterior to the atrio-ventricular groove. The relationship of the circumflex coronary artery maybe very difficult to determine and care should be taken not to transect it. An epicardial incision was used to open the aneurysm through its wall, which is usually very thin and friable. The aneurysm neck and sack were identified and closed.

Tissue samples of the aneurysm sacs were sent for histological study at the Histopathology Laboratories at the Universities of Cape Town and KwaZulu-Natal.

2. Results

2.1 Demographics [See Table 1]

There were 10 female and 10 male patients with a mean age of 17 ± 6 years (range 8 – 34 years). All presented with some degree of mitral regurgitation. There was equal gender distribution. Of the 20 patients four were of mixed race and 16 were of black African ethnic origin. Patients were referred from primary and secondary health institutions. The LVSMAs were diagnosed pre-operatively using transthoracic echo-cardiography and left ventriculography.

2.2 Symptoms [See Table 1]

Patients presented with a range of symptomatology, from asymptomatic (incidental findings) during routine examination to severe congestive cardiac failure and pulmonary edema. Four patients were diagnosed at their referral hospitals with incidental findings of mitral incompetence when they presented with unrelated complaints. Sixteen patients had clinical symptoms of exertional dyspnea, orthopnea and paroxysmal nocturnal dyspnea. Of the 16 patients presenting with symptoms and signs of cardiac failure, seven were in New York Heart Association (NYHA) Class II-III. Six patients were in NYHA Class III and four were in NYHA Class IV. Two patients presented in acute pulmonary edema and needed admission to the Intensive Care Unit (ICU). On further investigation by the referral hospital and at the tertiary care facilities, the diagnosis of LVSMAs was confirmed.

SUBMITRAL ANEURYSMS 1991 -- 2002						
CLINICAL DATA						
Patient	Age	Gender	Race	Presentation	Etiology and/or co-existent pathology	NYHA
M	31	F	B	4+ Mitral Incompetence, marked clinical symptoms	Congenital Submitral Aneurysm	4
T	23	F	B	Short History of Grade 2-3 dyspnea with 2+ Mitral Incompetence	Rheumatic Heart Disease	2
M 1	19	F	B	Pulmonary edema and 4+ Mitral Incompetence	Congenital Submitral Aneurysm	4
P	10	F	B	Grade 2 -3 dyspnea and 3+ Mitral Incompetence	Tuberculosis	2 - 3
M 3	8	M	C	Grade 2 -3 dyspnea and 3+ Mitral Incompetence	Tuberculosis	2 - 3
K	9	F	C	Grade 2 -3 dyspnea and 3+ Mitral Incompetence	Congenital Submitral Aneurysm	3
IM	12	F	B	Incidental finding, 1+ Mitral Incompetence	Congenital Submitral Aneurysm	1
IL	11	F	C	Short History of rapid deterioration and admitted in Pulmonary Edema. 4+ Mitral Incompetence	Congenital Submitral Aneurysm	4
K1	9	M	C	Documented Rheumatic fever and presented with Mitral and Aortic Incompetence	Rheumatic Heart Disease	3
IM 2	31	F	B	Incidental finding, 1+ Mitral Incompetence	No Histology	1
M	12	M	B	Grade 3 dyspnea, 3+ Mitral Incompetence	Congenital Submitral Aneurysm	3
M	16	F	B	Incidental finding, 1+ Mitral Incompetence	No Histology	1
K	22	M	B	Grade 3 dyspnea and 3+ Mitral incompetence	Infective Endocarditis	3
N	30	F	B	Grade 2 -3 dyspnea and 2+ Mitral Incompetence	Congenital Submitral Aneurysm	2
K2	5	M	B	Grade 2 -3 dyspnea and 2+ Mitral Incompetence	Infective Endocarditis	2
M	28	M	B	Previous Mitral valve replacement, decreased effort tolerance	No Histology	3
N	10	F	B	Incidental finding, 1+ Mitral Incompetence	Congenital Submitral Aneurysm	1
D	34	M	B	Grade 2 -3 dyspnea and 2+ Mitral Incompetence	Tuberculosis	2
H	32	M	B	Pulmonary Edema and 4+ Mitral Incompetence	Tuberculosis	4
B	15	M	B	Grade 3 dyspnea and 3+ Mitral incompetence	Congenital Submitral Aneurysm	3

2.3 Surgical Findings [See Table 2,3]

Patients were divided into three groups according to the extent of posterior mitral annulus involvement by the aneurysm neck at surgery:

Group I, single localized neck (n=12)

Group II, multiple necks (n=3)

(separate distinct openings, as opposed to pseudo division by the sub-mitral chordal apparatus)

Group III, involvement of the entire posterior mitral annulus (n=5)

Complete involvement of the posterior annulus has only recently been described in the literature by Du Toit et al [10]

The technique used to repair the LV SMA were as follows:

Totally intra-cardiac approach in 15 patients,

Totally extra-cardiac approach in two patients,

Combination of the two approaches in seven patients.

This breakdown includes:

Four re-operations in four patients 1 - 17 months after the initial operation

Two intra-operative failures of mitral valve repair.

Multiple different surgeons performed the surgery in the three different institutions.

Table 2

Surgical Findings

	Incidence	Surgical Approach	Closure of aneurysm	Repeat Surgery
Group I	60%			16%
12 Patients		Intra-cardiac - 9	64% Direct Suture - 43%	
14 Operations		Extra-cardiac - 2	15% Pericardial Patch - 57%	
2 Re-operations		Combined - 3	21%	
Group II	15%			
3 Patients		Intra-cardiac - 3	66% Pericardial Patch - 60%	
5 Operations		Combined - 2	33% Direct Suture - 40%	40%
2 Re-operations				
Group III	25%			
5 Patients		Intra-cardiac - 3	66% Pericardial Patch - 60%	0%
5 Operations		Combined - 2	33% Direct Suture - 40%	

The mean age of patients in Group III were 29.0 ± 5.1 years and was substantially older than the other groups; Group I - 12.2 ± 4.4 years, Group II - 10.3 ± 0.4 years

The most common technique used to close the neck of the aneurysms (inclusive of re-operations) was a primary patch closure with bovine or autologous pericardium (n=14). Direct suture closure was used with pledgeted sutures incorporating the aneurysm neck into repair/replacement of the mitral valve (n=10). In the five patients in Group III, aneurysms involving the entire posterior mitral annulus, direct suture was used in two and pericardial patch was used in three patients.

The mitral valve was repaired in 12 patients, seven in Group I, three in Group II and two in Group III. Valve replacement was necessary in six patients as a primary method of dealing with the mitral valve. In the extra-cardiac repairs the mitral valve was normal. In one patient in Group I and one patient in Group III, a mitral valve repair was initially attempted but abandoned intra-operatively, because of severe distortion of the subvalvar mitral apparatus following closure of the submitral aneurysm. Mitral valve repair was found to be inadequate postoperatively at follow up in a further four patients (20%), all of whom also had failure of closure of the LV aneurysm neck. There were thus two intra-operative and four post-operative failures of the mitral valve repair.

Failure of closure of the sub-mitral left ventricular aneurysm, necessitating a repeat surgical procedure occurred in four patients (20% failure rate) – two with Type I defects (16%), and two with Type II defects (66%). In the patients with Type II defects it was found that they had additional aneurysm necks, which had not been initially identified. These were noted to be separate distinct openings and not pseudo division by the submitral apparatus. This finding could possibly explain the high failure rate in Type II defects. Echocardiographic examination had revealed recurrent enlargement of the LV aneurysm and progressive mitral regurgitation. The initial surgical approach had been a combined approach in three of the four patients needing repeat procedures, more probably reflecting the complexity initially faced as opposed to the approach per se. In three

patients the initial closure technique had been by direct suture versus one patient in whom a patch had been initially used. Re-repair was done 1–17 months after the initial surgery (average of 6.3 ± 7.5 months). Pericardial patch closure of the aneurysm was used in all the patients coming for re-repair of their aneurysms.

There was no operative mortality and all patients were discharged from the hospital in NYHA Class I

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SURGICAL FINDINGS AND RESULTS

Group	Patient	Age	Gender	Pre Operative Mitral Regurgitation	NYHA	Mode of Failure	Surgical approach	LV aneurysm closure	Mitral valve
I	BM	16	F	1+	1		Combined	Direct suture	Repair – including Flexible ring
I	JK	9	F	3+	3		Intra-cardiac	Direct suture	Repair - including Rigid Ring
	JK-Reop			3+	2	Failure of LVMA closure and Mitral valve repair	Intra-cardiac	Pericardial patch	Failure of Mitral valve repair: Replacement
I	LM	12	M	3+	3		Combined	Direct suture	Mitral valve replacement
I	MM	12	F	1+	1		Intra-cardiac	Direct suture	Repair - including Rigid Ring
I	NM 1	19	F	4+	4		Intra-cardiac	Direct suture	Repair – Modified Kay annuloplasty
I	NM 3	8	M	3+	2-3		Combined	Pericardial patch	Repair - including Rigid Ring
	NM 3-Reop			3+	2-3	Failure of LVMA closure and Mitral valve repair	Intra-cardiac	Pericardial patch	Failure of Mitral valve repair: Replacement
I	TK	22	M	3+	2	Intra op Mitral valve repair failure	Intra-cardiac	Pericardial patch	Repair - including Rigid Ring but intr op assessment poor and replacement done
I	KN	30	F	2+	2		Intra-cardiac	Pericardial patch	Repair -Plication of posterior leaflet, including Rigid Ring
I	RK2	5	M	2+	2		Extra-cardiac	Pericardial patch	No Mitral surgery
I	SM	48	M	Previous Mitral replacement	2		Intra-cardiac	Pericardial patch	Re-do Mitral valve replacement
I	MN	10	F	1+	1		Extra-cardiac	Pericardial patch	No Mitral surgery
I	RK1	9	M	3+ MR, 3+ AR	3		Intra-cardiac	Direct suture	Replacement, Mitral and Aortic valve
II	MD	34	M	2+	2		Intra-cardiac	Pericardial patch	Repair – including Flexible ring
II	NL	11	F	4+	4		Combined	Direct suture	Repair - including Rigid ring
	NL-Reop			0	1	Failure of closure of LV aneurysm	Intra-cardiac	Pericardial patch	Re-assessment - No further surgery
II	TP	9	F	3+	2-3		Combined	Direct suture	Repair - including Rigid Ring
	TP-Reop			0	2-3	Failure of closure of LV aneurysm	Intra-cardiac	Pericardial patch	Re-assessment - No further surgery
III	BT	23	F	2+	2		Combined	Patch repair	Mitral valve replacement
III	JM	31	F	4+	4	Intra op Mitral valve repair failure	Intra-cardiac	Direct suture	Repair initially attempted but valve replacement done
III	NM 2	31	F	1+	1		Intra-cardiac	Direct suture	Repair – including Flexible ring
III	MH	32	M	3+	4		Intra-cardiac	Patch repair	Mitral valve replacement
III	MB	15	M	3+ MR, 1+ AR	3		Combined	Patch repair	Mitral valve replacement

2.3 Laboratory findings [See Table 3]

In two patients with clinical rheumatic disease the Antistreptolysin O titre (ASOT) were raised above 200 IU and they both had histological findings suggestive of associated active rheumatic carditis. In four patients with clinical and microbiological evidence of tuberculosis the histological evidence confirmed active tuberculosis. Pre-operative blood cultures done suggested concomitant infective endocarditis in two patients, this was also confirmed with histology.

Of the remaining 12 patients no specific etiology was identified in nine patients leaving a presumed diagnosis of congenital submitral aneurysm.

Thus based on histopathology and laboratory investigations co-existent pathology was thought to be:

1. Rheumatic heart disease - 2 patients
2. Tuberculosis - 4 patients
3. Infective endocarditis - 2 patients

Unfortunately no histology was obtained in three patients

Table 3					HISTOLOGY OF SUBMITRAL ANEURYSMS				
Patient	Age	Gender	Findings			Etiology and associated pathology			
JM	31	F	Vascularised fibrous tissue forming the wall of the aneurysm. Inner lining contains some fibrin. No active of chronic inflammation seen. Features consistent with Congenital Submitral Aneurysm						
NM 1	19	F	Neovascularisation with several fibroblasts and poorly preserved Aschoff bodies. Features consistent with active Rheumatic Heart Disease			Rheumatic Heart Disease			
BT	23	F	Section of aneurysm wall consisting of fibrous tissue scanty hypertrophied myocytes, chronic non specific inflammation, foam cells and thick walled vessels consistent Congenital Submitral Aneurysm						
TP	10	F	Areas of fibrosis and chronic inflammation with caseating granulomatous necrosis.			Tuberculosis			
NM 3	8	M	Thin wall aneurysm with areas of granulation tissue repair. Within the wall there focal granulomas with caseation.			Tuberculosis			
JK	9	F	Large aggregates of amorphous degenerate collagen consistent with fibrinoid necrosis and collagen degeneration.						
MM	12	F	Large aggregates of fibrinoid material and degenerate collagen.						
NL	11	F	Large aggregates of fibrinoid material and degenerate collagen.						
RK	9	M	Neovascularisation, fibroblasts and poorly preserved Aschoff bodies.			Rheumatic Heart Disease			
NM 2	31	F	No Histology						
LM	12	M	Large aggregates of fibrinoid material and degenerate collagen, chronic non specific inflammation			Congenital Submitral Aneurysm			
BM	16	F	No Histology						
TK	22	M	Section of aneurysm wall consistent with features of active infection with degranulating macrophages.			Infective Endocarditis			
KN	30	F	Fibrous tissue scanty hypertrophied myocytes, chronic non specific inflammation.						
RK	5	M	Features of active infection with degranulating macrophages.			Infective Endocarditis			
SM	28	M	No Histology						
MN	10	F	Vascularised fibrous tissue forming the wall of the aneurysm. Inner lining contains some fibrin. No active of chronic inflammation seen.						
MD	34	M	Areas of granulation tissue focal granulomas and no caseation. Ziehl Nielsen stain +			Tuberculosis			
MH	32	M	Granulation tissue, focal granulomas and caseation.			Tuberculosis			
MB	15	M	Large aggregates of fibrinoid material and degenerate collagen.						

2.4 Diagnostic Investigations

In examining the chest roentgenograms of the patients in this series it was found that that an abnormal cardiac contour was absent in 15% (3 out of 20 patients) of chest roentgenograms. The remainder of the patients had left atrial enlargement and generalized, somewhat nonspecific, cardiac enlargement. The lung fields demonstrated upper lobe diversion of pulmonary blood flow consistent with pulmonary venous congestion secondary to mitral incompetence as found in 85% (17 out of 20 patients) Calcification is said to be useful and very important diagnostic feature, but will be present only if the aneurysm is not of recent origin.(1*) In this series, however, there were no calcified aneurysms seen.

2.5 Follow up

Follow up data was obtained in 15 of the patients discharged (75 %) with a follow-up range of 1 – 48 months. The low percentage of follow-up are a result of poor socio-economic and other demographic factors, with many of our patients returning to rural areas with no mechanism of follow-up. Of the 15 patients followed, there were 10 mitral valve repairs and 5 valve replacements, none had mitral regurgitation or aneurysm recurrence.

DISCUSSION

Epidemiology

The LVSMA is a rare entity that occurs predominantly in young patients of black African ethnic origin, almost exclusively from poor socio-economic groups in sub-Saharan Africa (countries with one of the highest incidence of tuberculosis and rheumatic fever in the world), the southern part of India and recently Brazil. There have also been reports from Europe, North America, Japan and Australia (mainly in Oriental and black African immigrants), [1-8].

Twenty patients in almost as many years were seen in three institutions in a country with one of the highest incidences of LVSMA.

Aetiology

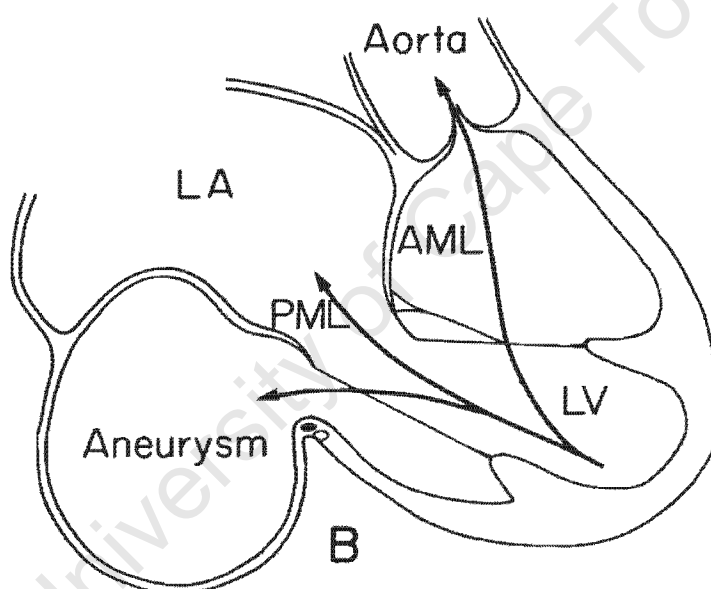
The aetiology of submitral ventricular aneurysms remains uncertain. The evidence accumulated in this series and a review of the available reports in the literature supports the hypothesis of a congenital or developmental aetiology. In addition to this, this series identified co-existent inflammatory pathology (rheumatic carditis, tuberculosis and infective endocarditis) on histology in 40% of the patients.

1. Because of the predilection for the same anatomical sites and the absence of evidence of other causes in many cases, (such as atherosclerotic coronary artery diseases, syphilis, bacterial endocarditis, tuberculosis and trauma), Abrahams and co-workers [1], and others [3,4,5,6,9,10,14], suggested a congenital or developmental aetiology. This postulate was also supported by Chesler [2], who stated that:

"a defect in the muscular fibrous junction below the intermediate portion of the left aortic cusp or the posterior

Clinical Presentation

Mitral regurgitation is present in most patients [1-10] and may range from trivial to severe. Mitral regurgitation results from dilatation or distortion of the posterior annulus, with loss of support of the posterior leaflet at its base and failure of leaflet co-aptation. With increasing dilatation or distortion of the ring, chordae tendinae stretch or rupture with the production of a prolapsed or flail leaflet and gross mitral incompetence. (Figure 1)



(Figure 1): Diagram demonstrating loss of leaflet co-aptation due to the effect of the aneurysm on the Mitral Valve. (AL – Anterior mitral leaflet, PL – Posterior mitral leaflet)

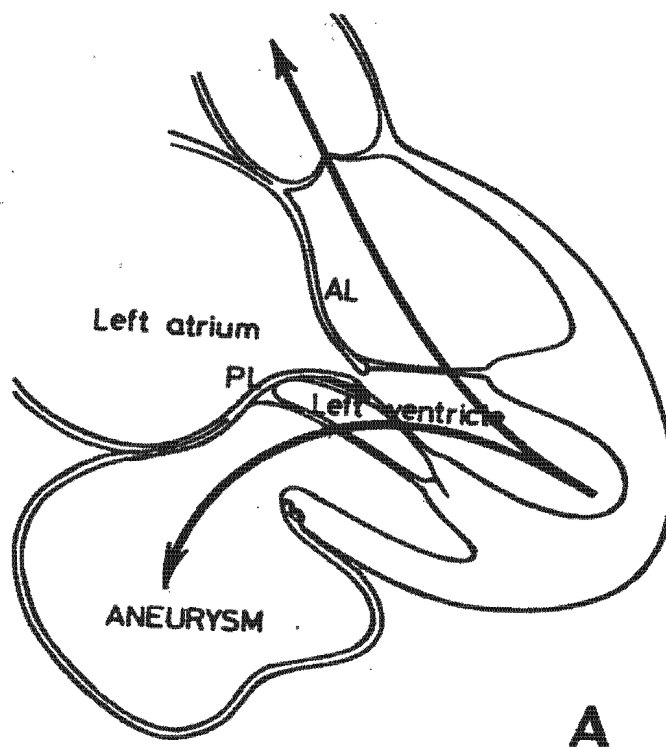
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 Perspectives on the Mitral valve
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Clinical features of submitral aneurysms may include systemic embolism [1,5], angina pectoris [2,5,7,9], myocardial infarction [5,7,9], supra-ventricular and ventricular tachy-arrhythmias [2,4], and cardiac failure [5,1*]. The condition may also mimic rheumatic carditis [9,17,20] or dilated cardiomyopathy [1*]. Coronary arteriography may reveal circumflex coronary artery compression or stretching especially resulting in angina pectoris [13][Fig 7].

In areas such as Southern Africa, where rheumatic carditis and dilated cardiomyopathy are common causes of heart disease in the ethnic African population, the diagnosis of a submitral aneurysm may be missed [1*].

On examination the character of the left ventricular impulse is often of value in differentiating between cardiomyopathy and a submitral aneurysm. Unlike the impulse of the dilated poor myocardium in idiopathic cardiomyopathy the apex beat is forceful owing to hypertrophy of a good myocardium in response to the diastolic overload produced by the aneurysm [1*].

In many cases a palpable impulse is present above the apex and along the left cardiac border. This impulse has a "double kick" and is a result of the aneurysm expanding in late systole. Because these aneurysms are posterior to the heart, the palpable double impulse is often a referred pulsation analogous to a left atrial lift. (Figure 2)



(Figure 2): Diagram demonstrating cardiac enlargement and filling of the aneurysm sac .

(AL – Anterior mitral leaflet, PL – Posterior mitral leaflet)

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Perspectives on the Mitral valve

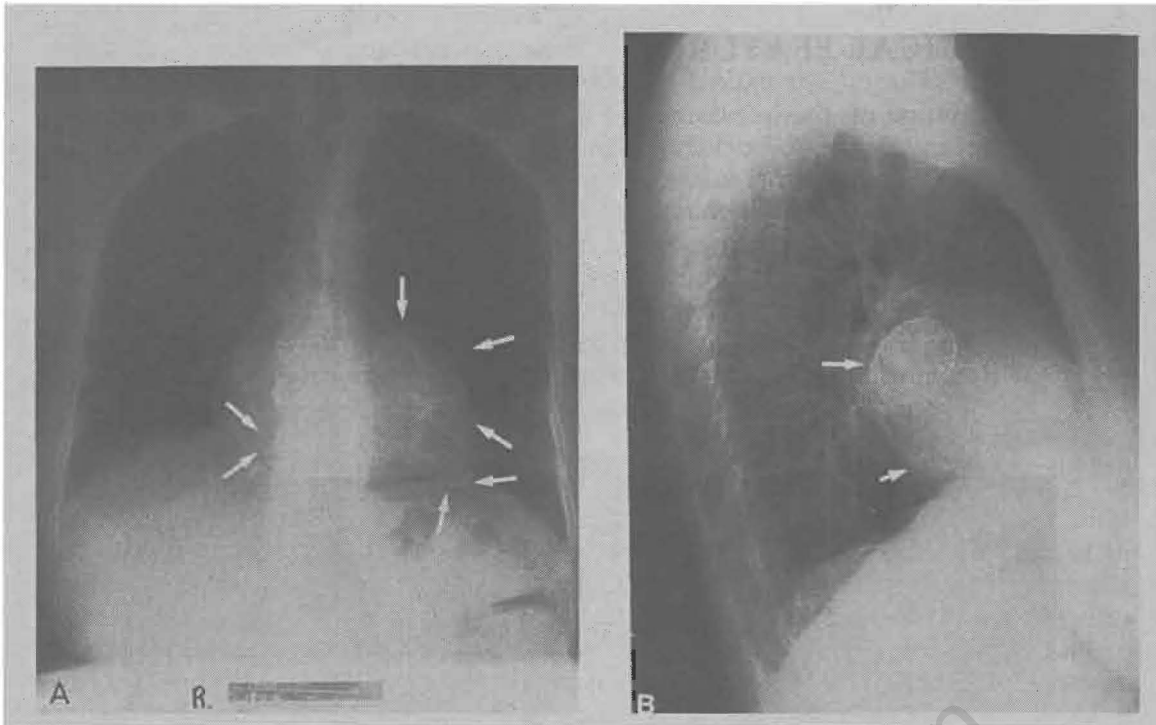
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Diagnosis

1. Radiological Features

Cockshott [11], in his review of 50 patients encountered in Nigeria, stated that "in most cases the diagnosis is readily established by plain radiography" However, other investigators, shows that an abnormal cardiac contour may be absent [1*]. It may also be masked by left atrial enlargement and generalized, somewhat nonspecific, cardiac enlargement. An abnormal bulge on the left border of the heart is a recognized feature of a LVSM [11,1*] In some patients, the aneurysm may be detected posterior to the cardiac silhouette in a lateral view. Calcification is a useful and very important diagnostic feature, but will be

present only if the aneurysm is not of recent origin [14,1*]. The lung fields may commonly demonstrate upper lobe diversion of pulmonary blood flow consistent with pulmonary venous congestion.



(Figure 3) Postero-anterior (A) and lateral (B) chest radiograph of a 64 year old woman, showing large multiloculated calcified submitral aneurysm. (Arrows)

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2. Electrocardiographical Features

The ECG is normal in a minority of cases, and Chessler found only one completely normal ECG in 15 patients with submitral aneurysms [2].

Electrocardiographical evidence of left ventricular hypertrophy, out of proportion to the severity of the mitral regurgitation, may suggest the possibility of an additional hemodynamic load on the left ventricle [1*]. Low voltages, nonspecific ST-segment and T-wave changes and signs of myocardial ischaemia or infarction may be encountered. Electrocardiographical evidence of ischaemia is often due to distortion or obstruction of the left circumflex coronary artery along its course in the atrioventricular groove [4,9,13]. Patients presenting with ventricular tachycardia have also been encountered [4]. Atrial fibrillation is also common due to the left atrial enlargement [14].

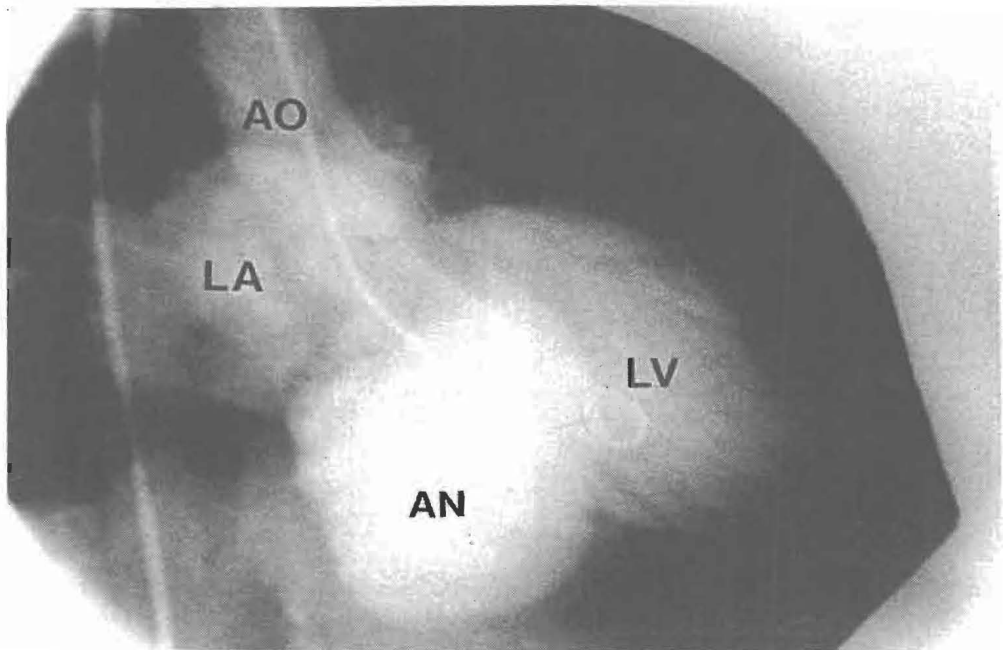
Conduction disturbances such as first-degree heart block are not uncommon and may also be related to myocardial ischaemia produced by

pressure on a coronary artery [13]. Unfortunately the electrocardiograms of patients in this series were not all available.

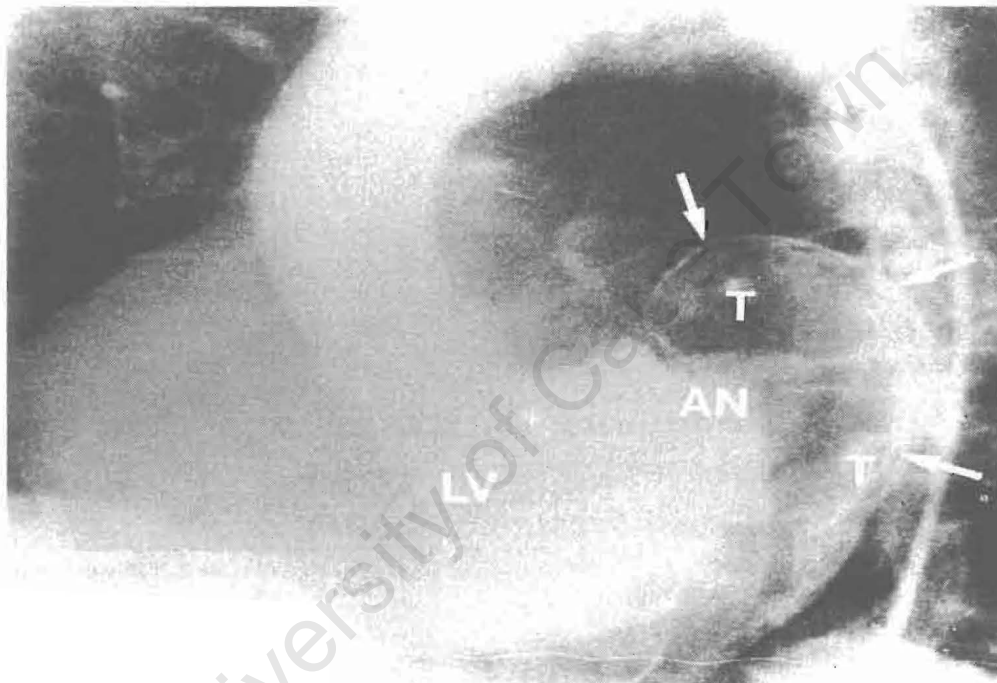
3. Cardiac Catheterization and Angiocardiography

Until the advent of echocardiography and computerized tomography (CT), cardiac catheterization and angiocardiography were necessary to confirm the diagnosis, locate the origin of the aneurysm, and assess the severity of the hemodynamic disturbance. Left ventriculography in the right anterior oblique position usually demonstrates the aneurysm, but multiple views may be required. Unless the diagnosis is suspected before the study is undertaken the aneurysm may be overlooked, particularly if it is small and mitral regurgitation is present. Large aneurysms may displace the cardiac shadow. In such instances, a left ventriculogram in the postero-anterior position is necessary to assess the position of the heart. In other cases, the left atrium may be displaced superiorly and can be considerably compressed. An aneurysm may only partially fill with dye, or may not fill at all, if it contains thrombus. The catheter may also enter a wide-necked aneurysm, which can then be outlined by direct injection of contrast medium. (Figure 4 A,B)

A



B



(Figure 4 A, B) (A) Angiogram in the right anterior oblique position. (B) Angiogram in the Left lateral position, both demonstrating a LV SMA. (AO) Aorta, (LV) Left ventricle, (AN) Aneurysm, (LA) Left atrium, (T) Thoracic aorta. The arrows in (B) demonstrates the calcified rim of the LV SMA

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4. Echocardiographic Features

2-D echocardiography demonstrates a submitral aneurysm as an echo-free space arising below the posterior mitral leaflet extending postero-medially and superiorly to compress the left atrial cavity. Communication with the left ventricular cavity can then be seen in the region of the atrioventricular groove. The mitral valve apparatus is very well visualized and mitral incompetence can readily be demonstrated. Mitral incompetence is usually due to lack of leaflet co-aptation and distortion of the mitral valve. Because of cardiac displacement and variable position of the aneurysms, unconventional views are sometimes necessary to demonstrate them. Left ventricular (LV) contrast echocardiography can also be used [23]. A pigtail catheter is placed in the LV and the patients blood is used as the contrast agent to determine mitral regurgitation and to locate the neck of an aneurysm. A flail posterior leaflet provides important ancillary evidence of the existence of a submitral aneurysm, provided that other causes such as degenerative myxomatous disease, trauma, infective endocarditis, and acute rheumatic carditis have been excluded.

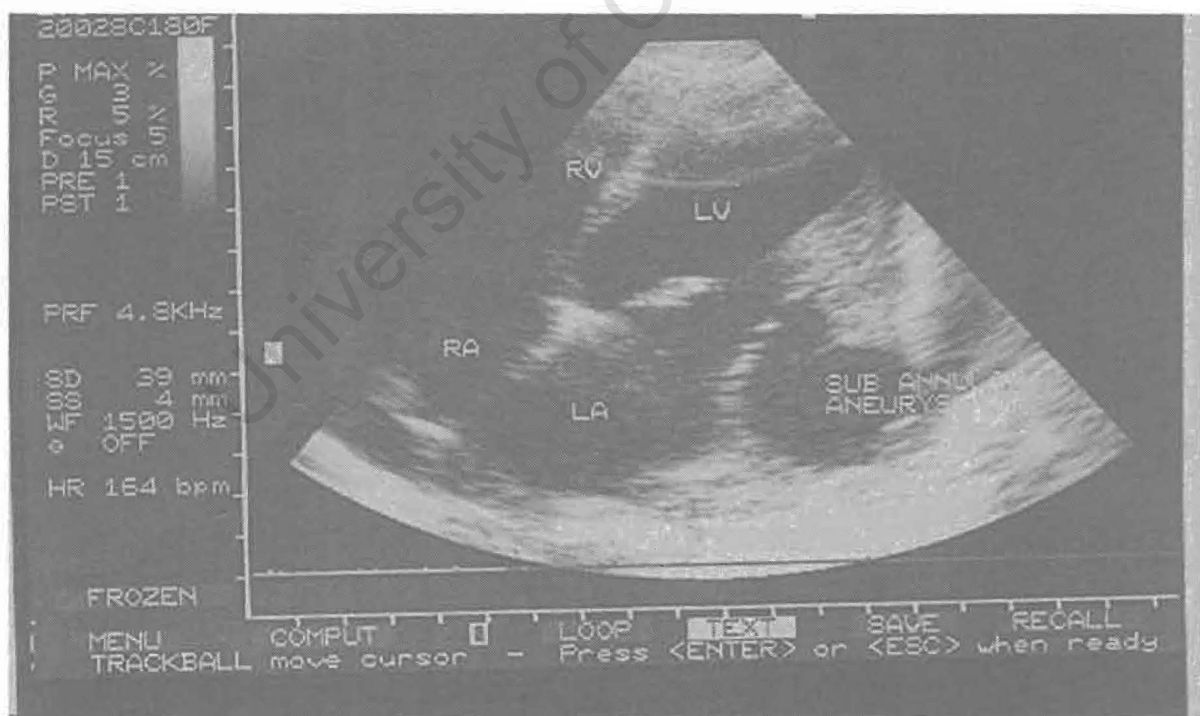


Fig 5 A

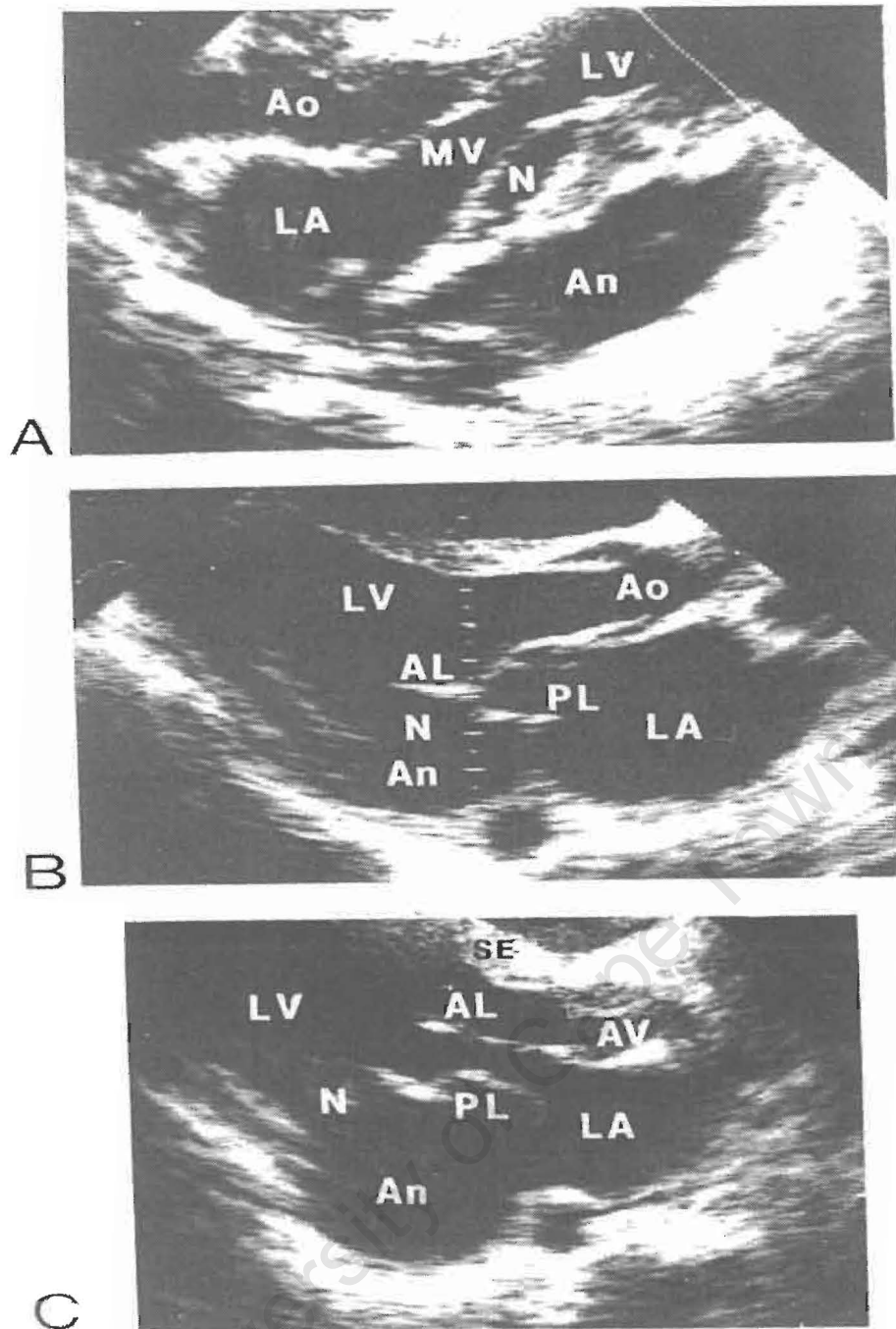


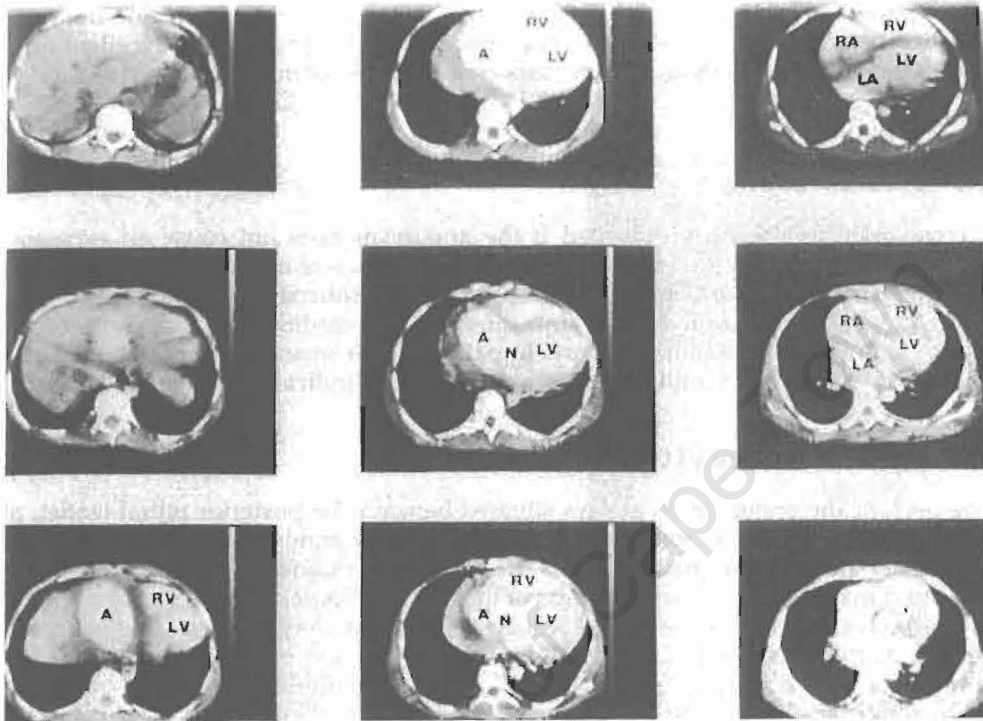
Fig 5 B

(Figure 5 A,B) 2-D echocardiography pictures demonstrating two different patients with LV SMA. The LV SMA is seen as an echo-free space arising below the posterior mitral leaflet extending postero-medially and superiorly to compress the left atrial cavity. (Ao) Aorta, (LA) Left atrium, (MV) Mitral valve, (An) Aneurysm, (LV) Left ventricle, (N) Neck of aneurysm, (AL) Anterior leaflet on mitral valve, (PL) Posterior leaflet of mitral valve

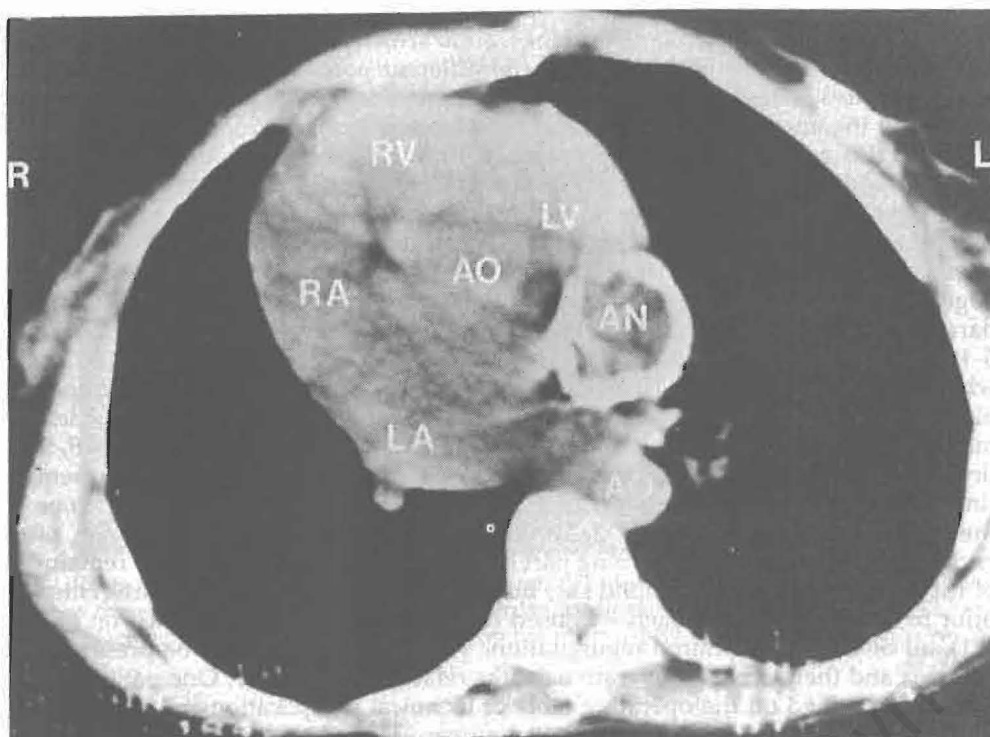
5. Computed Tomography (CT)

The use of CT and contrast CT in defining the position, size, presence and extent of thrombus and calcification has represented a major advance in the diagnosis and assessment of submitral aneurysms. Barlow reports that CT was positive in all seven patients in whom it was performed, and the information obtained equaled or surpassed that derived from angiography [1*]. CT provides an excellent non-invasive method of investigation in suspected cases of submitral aneurysm.

A



B



(Figure 6) A) Consecutive contrasted Computed tomography (CT) scan in a 31 year old male demonstrating the (A) aneurysm and its communication, via a wide neck (N) with the left ventricle (LV) Extensive thrombus formation is seen in the aneurysm.

(Figure 6) B) Single frame of CT scan sequence clearly demonstrating the LV SMA (AN) in relation to the aorta (AO), left ventricle (LV) and the left atrium (LA).

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Natural History

Death is sometimes sudden, the diagnosis being made at necropsy. This may be due to acute pulmonary edema, rupture of the aneurysm [1,14] or an acute arrhythmia [4]. In most patients however, a severe work overload on the heart is produced by the aneurysm or by valvular incompetence. This results in cardiac failure [2,3,8,14]. Associated myocardial ischaemia as a result of circumflex artery occlusion [12], clot dislodgement with cerebral embolism [1] have been described.

Interestingly, Cockshott [11] divided his patients into two groups:

Group I -- Those in whom LVSMA was the cause of significant presenting symptoms.

Group II -- Those in whom LVSMA was an incidental finding with few symptoms.

Investigation by Cockshott of his patients in Group II, showed partial or total obliteration of the cavity by thrombus, or a small aneurysm cavity. He observed marked "spontaneous" improvement in several, presumably a result of partial or complete thrombosis in the aneurysm.

In contrast to the findings of Cockshott of his patients in Group II, the observation in this series that patients with complete annular involvement comprised an older group would support a hypothesis that involvement of the sub-mitral annulus may be progressive. This series and others [8,10,19] have shown the progressive nature of the disease as opposed to Cockshott. Conclusive evidence of the natural history for LVSMA lacks, although this series postulates that these aneurysms could progress and involve the whole posterior annulus. Progression to intractable cardiac failure if these aneurysms are not treated surgically is very likely.

In support of the congenital developmental theory, according to Wolpowitz, it is notable that the part of the left ventricular wall at the atrioventricular junction is unlike that of the rest of the left ventricle in the hearts of young embryos [8]. At an early stage of development, the wall of the left ventricle has only a thin cortical layer of muscle, and thereafter its increase in thickness is due to an irregular thickening that becomes sponge-like. Most of these spaces are then obliterated by the continued growth of the primitive myocardium, except for the area of the ventricle underlying the posterior cusp of the mitral valve, which persists as a cleft well into fetal life, and sometimes even in the full term fetus. The sequence of events is possibly related to an existing defect in the ventricular wall that is enlarged by the intra-ventricular tension developed during isometric contraction of the left ventricle. This would support the hypothesis that the LV SMA is a congenital pulsion diverticulum. Several ostia between the trabeculae carneae will then coalesce over time to form a larger opening and thus result in the neck of the aneurysm with multiple ostia or complete involvement of the posterior annulus.

The understanding in the co-existent "inflammatory aetiology" patients, is that a defect in the posterior annulus develops due to the localized effects of the inflammatory process with resultant weakening of the annulus. The resultant progression of the aneurysm is then related to the defect in the ventricular wall that is enlarged by the intra-ventricular tension developed during isometric contraction of the left ventricle.

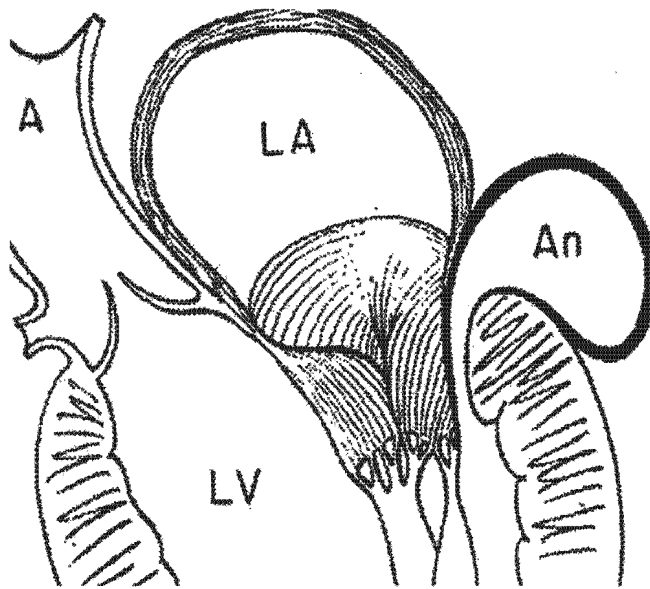
Regardless of the etiology, cardiac enlargement is the rule, partly because of the size of the aneurysm, but also because of the associated hemodynamic effects of an aneurysm distorting the mitral valve. This impairs the function of the left ventricle and mitral valve apparatus resulting in mitral regurgitation. Progressive mitral incompetence exacerbates the left sided cardiac failure, producing pulmonary hypertension with resultant right sided ventricular failure.

1. Histology

The typical wall of "congenital" aneurysms consists of laminated thrombus, avascular fibrous tissue that contains chronic inflammatory cells with scattered deposits of hemosiderin, degenerate collagen and very scanty compressed muscle fibers. Areas of fibrinoid necrosis is common. Tuberculous myocarditis is diagnosed by demonstrating Langhans granulomatous cells and/or caseation in the wall of the aneurysm. The finding of Aschoff bodies in the aneurysm wall confirms rheumatic heart disease. The diagnosis of infective endocarditis is made by demonstrating organisms in serial blood cultures and supported by active degranulating macrophages on histological examination.

2. Pathological Anatomy

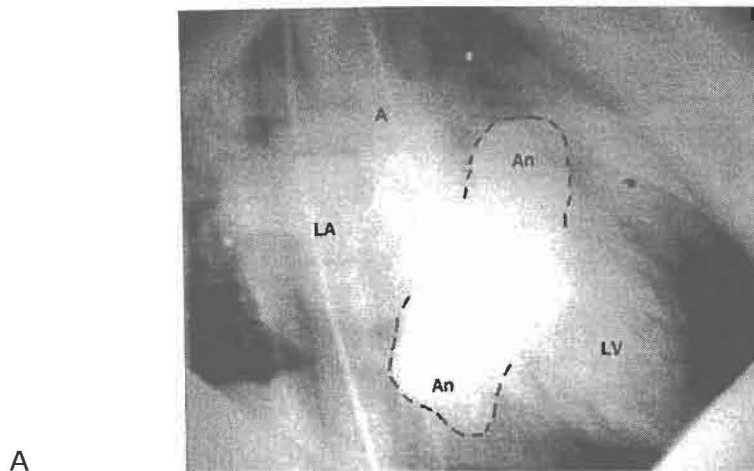
The neck of the aneurysm is always situated beneath the posterior mitral leaflet, at any site between the antero-lateral and postero-medial commissures, although it more commonly arises at the medial half of the annulus. The transverse diameter is variable, but it may be up to two thirds of the annulus. Consequently, a large segment of the posterior leaflet has no annular attachment. As the neck may be circular or oval, the large vertical diameter may give a false impression of a deficiency in the left ventricular posterior wall. The unattached posterior mitral leaflet usually forms part of the roof of the aneurysm, which is contiguous with the floor of the left atrium. (Figure 7)



(Figure 7): Diagram demonstrating the position of the aneurysm in relation to the mitral valve and the left atrium (A - Aorta, LA - Left Atrium, AN - Aneurysm, LV - Left ventricle)

From : Abrahams DG , Barton CJ , Cockshot WP.
Annular Subvalvar left ventricular aneurysms
*Q J Med*1962;31:345-360

The direction of expansion of the aneurysm is extremely variable and influences the function of the mitral valve. In our experience, there is usually some protrusion into the left atrial cavity, which results in the posterior leaflet being pulled posteriorly, causing valve regurgitation. Occasionally, the aneurysm may perforate into the left atrial cavity. Most of the aneurysmal sac lies behind the left ventricular posterior wall to which it is usually closely adherent. The cavity is often multi-loculated and the different portions communicate through narrow orifices. These may be confused with the true neck of the aneurysm during surgery, if the aneurysm is approached from outside the heart. The aneurysms may have single or multiple necks, and in some cases, the neck may involve the whole of the posterior annulus as seen in the following angiograms. (Figure 8 A,B)



(Figure 8 A): Left Ventriculograms in the right anterior oblique plane. LV SMA expanded and involving the whole mitral annulus as found in two of the patients in Group III. Contrast medium has been injected into the left ventricle (LV) via the aortic root (A). The contrast media is seen filling the LV and the left atrium (LA) indicating severe mitral incompetence. The LV SMA (AN) is outlined with the dashed line.

Fig A: With Permission: H.J. Du Toit et al.

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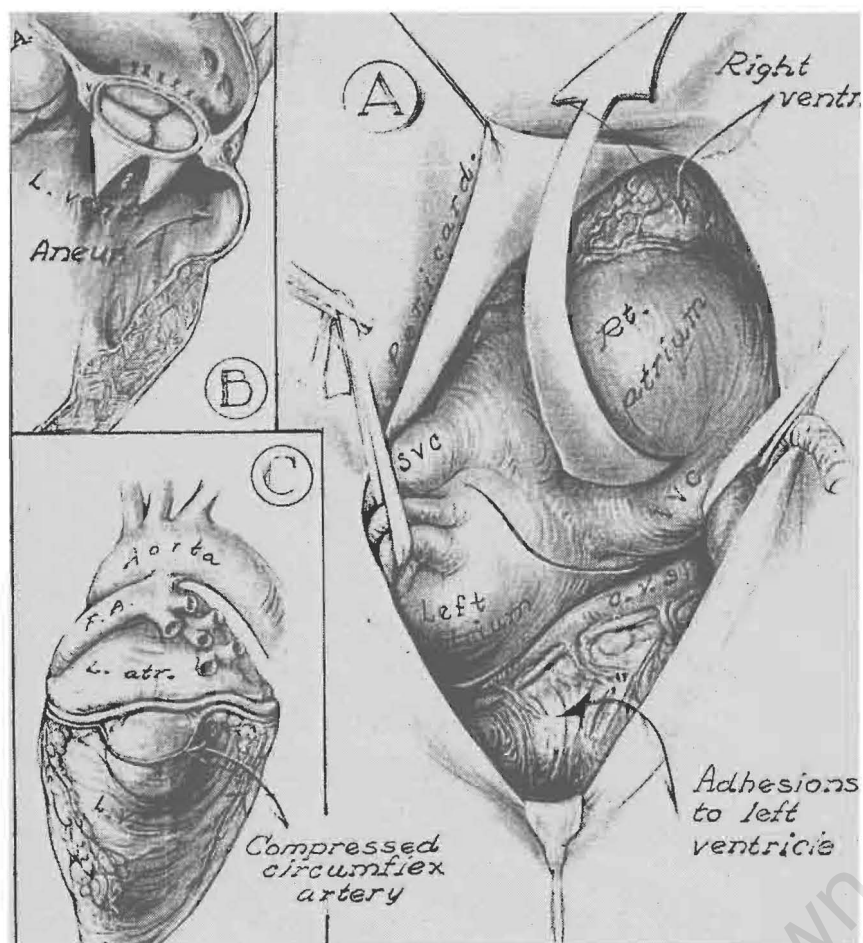
Aneurysms tend to be large as shown in the intra-operative photograph because they track in a circular direction. Enlargement is laterally, anteriorly and superiorly by herniation through the attachment of the left ventricular muscle and the fibrous valve ring. They may be loculated and in all instances contain clot, fresh and laminated, as well as cellular debris. Calcification of the laminated clot may develop (Figure 9)



(Figure 9): Intra-operative photograph of a LVSM, shown here as the whitish bulging structure to the left of the image. The aortic cannula (cranial) is seen in the centre bottom of the photograph and the diaphragm at the top (caudal) Arrows indicating LVSM

The left atrium may be compressed and the circumflex artery may be greatly stretched and narrowed along its course in the atrio-ventricular groove by the aneurysm. This may produce symptoms of angina pectoris and may lead to myocardial infarction due to coronary artery occlusion or thrombosis.

(Figure 10,11)

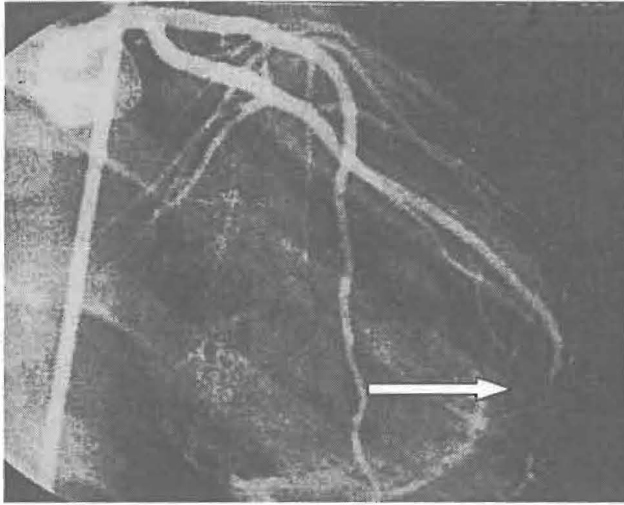


(Figure 10) Diagrammatic representation of stretching and compression of the circumflex coronary artery by the enlarging aneurysm. A) Reflection of the pericardium demonstrating the relationship of the cardiac structures. B) Saggital view of the LV SMA. C) Compression of the circumflex coronary artery by the aneurysm

From: Clerkin JP, Bunje H

Rare cardiac aneurysm in a young adult

Thorax 1955;10:42-45



(Figure 11) Angiogram of the Left coronary system in the right anterior oblique plane. Stretching and partial occlusion of the circumflex artery is demonstrated (lower right of picture)

From: Clerkin JP, Bunje H

Rare cardiac aneurysm in a young adult

Thorax 1955;10:42-45

University of Cape Town

1. Medical

Conservative treatment maybe indicated if the aneurysm does not cause an excessive hemodynamic load on the left ventricle by virtue of its size or the presence of severe mitral regurgitation. Medical management comprises anticoagulation to prevent systemic embolization, antibiotic prophylaxis against infective endocarditis in patients with mitral regurgitation, and anti-failure therapy. Evidence in this series and others [4,8,10,12,19,24] do however suggest that surgical intervention is the only definitive method of treatment in this condition. Medical management may be used as a temporizing method until an elective date can be determined for the patient or to improve the pre-operative state of patients who are in severe cardiac failure.

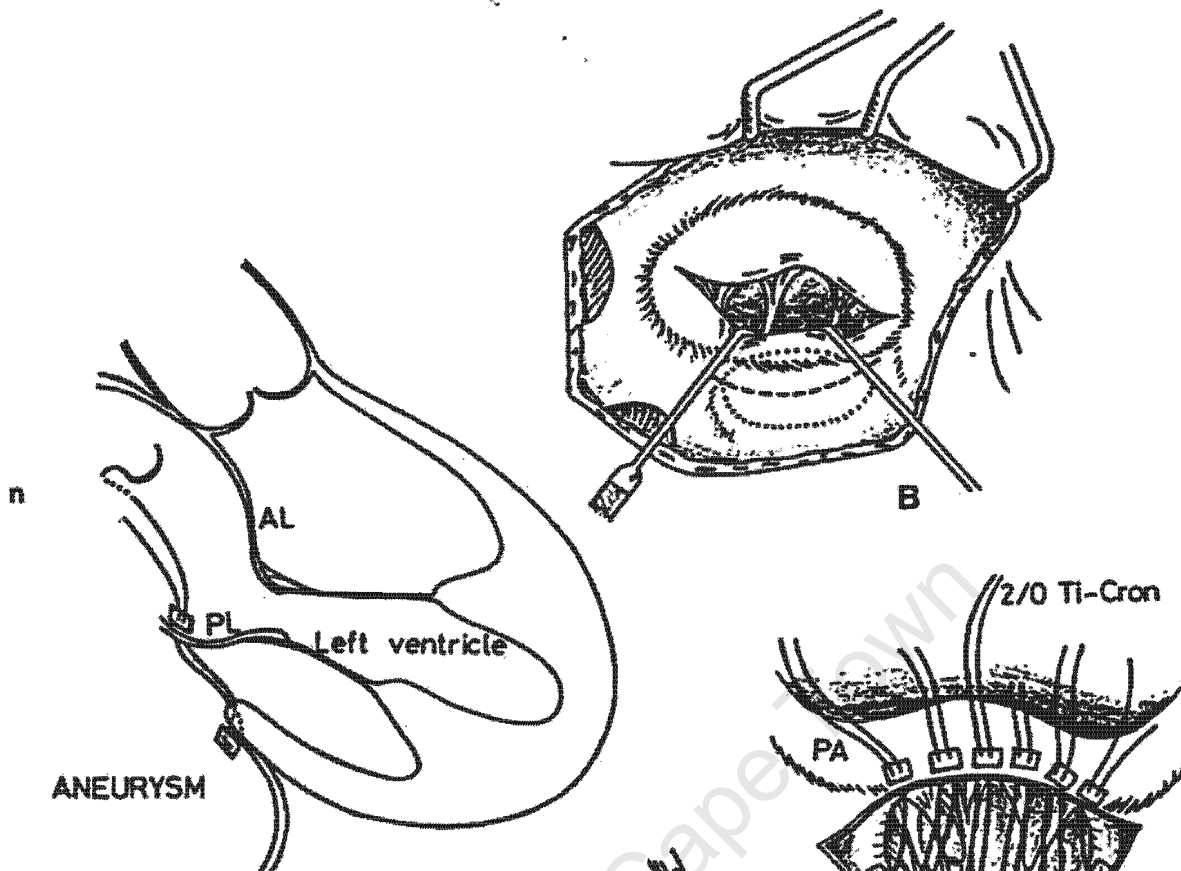
2. Surgical

Two techniques are described in the literature to approach and repair submitral aneurysms: an intra-cardiac approach and an extra-cardiac approach. In our experience these aneurysms could not be exclusively approached via one exclusive technique. Successful repair needs careful assessment and planning taking into consideration variables like extent of involvement of the posterior mitral annulus, aneurysm size, degree of mitral regurgitation and surgical experience.

i. Intracardiac Approach

This technique was described by Antunes [19]. Access to the left atrium is via and incision through the inter-atrial groove. The neck of the submitral aneurysm is identified by retracting the posterior mitral leaflet using valve hooks, and examining the subvalvar area between the mitral valve annulus and papillary muscles. A "right angled" instrument is then placed in the identified aneurysm to establish the relationship between the aneurysm, the left ventricular free wall and the posterior left atrial wall. A curved incision is then made in the floor of the left atrium and or posterior mitral leaflet parallel to the posterior mitral annulus to allow entry into the aneurysm sac. The mitral valve leaflets are usually normal. The advantage of this 'intra-cardiac' approach is that the mitral valve can be assessed and tested for competence following repair of the submitral aneurysm.

Limitations of this approach is, however, difficulty in exposing large complicated aneurysms. The aneurysms are so large that they displace and obscure the normal anatomy. In these cases either an extra-cardiac or a combination of an intra-cardiac and extra-cardiac approach should be used [12,19,24,25].



(Figure 12) Diagram demonstrating the intra-cardiac approach

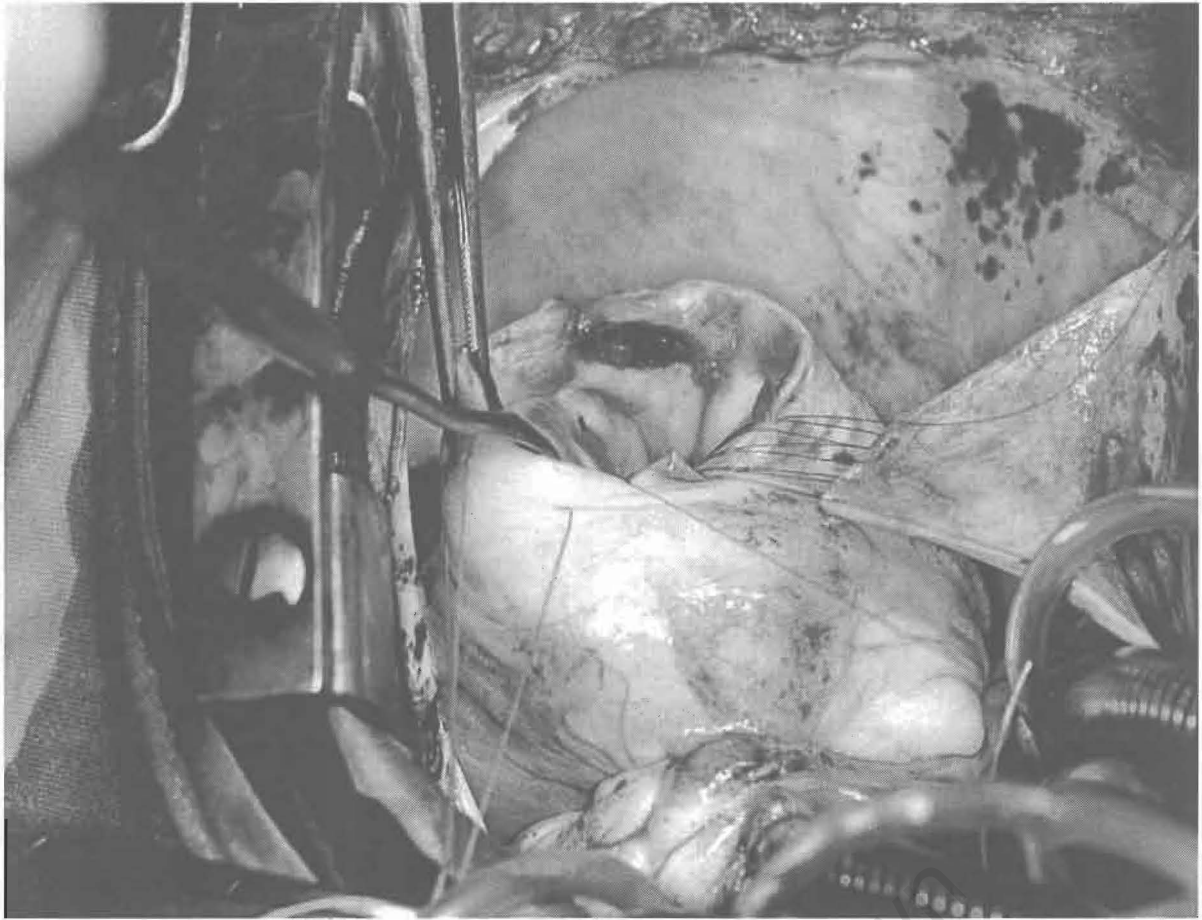
(Top) Exposure of the aneurysm neck below PML using a pair of valve hooks. (Left & Right bottom) Approximation of posterior mitral leaflet (PML) to left ventricular wall using 2-0 Ticron pledgeted sutures to close the neck of the aneurysm and re-approximate the posterior left ventricular free wall.

With permission:

MJ Antunes

Submitral Left Ventricular Aneurysms

J Thoracic Cardiovasc Surgery 1987;94:241-5



(Figure 13) Intra-operative photograph of extra-cardiac approach. The forceps and eyelid retractor are in the aneurysm cavity. A patch of autologous pericardium is being sewn into place

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Conclusions

In this series submitral aneurysms accounted for less than 1% of cases of isolated mitral valve lesions encountered.

The aetiology of LVSMA is generally considered to be a congenital weakness of the fibrous annulus of the valve, this is supported by the predominant single race group involved, as well as by the anatomy and embryology of the affected area.

This study identifies co-existent inflammatory or infective pathology in a third of patients.

Complete involvement of the entire posterior mitral annulus of patients in Group III suggests that there is a spectrum in this condition, single or multiple defects up to complete annular involvement.

The observation that patients with complete annular involvement comprised an older cohort of patients would support the hypothesis that involvement of the submitral annulus may be progressive.

The progressive nature of the disease also suggests that patients could eventually progress to intractable cardiac failure. Surgical intervention is the only method of treatment of LVSMA

Submitral aneurysms should not always be approached exclusively by one technique as the extent of involvement of the posterior mitral annulus, aneurysm size and degree of mitral valve incompetence varies greatly.

Surgical failure in this series was not related to the surgical approach, but rather due to failure in identifying additional aneurysm necks, inadequate closure of the aneurysm or failure of mitral valve repair.

Successful mitral valve repair is dependent on the appropriate understanding of the relationship between the aneurysm, the mitral valve

and the annulus. Patients with involvement of the entire posterior mitral valve annulus were more likely to require mitral valve replacement.

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