

BLOOD CYSTS OF THE CARDIAC VALVES IN ADULTS

Review and analysis of published cases

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Abstract

Background and aim of the study: Blood cysts of cardiac valves are generally seen in newborns and infants and very rarely in adults. Although in most cases they are incidental findings they may be associated to severe cardiac or systemic complications. This study analyzes incidence, presentation and treatment of valvular blood cysts in adults. Methods: A review of the pertinent literature through a search mainly on PubMed and Medline was performed. Results: In patients [?] 18 years of age, our search disclosed 54 patients with mitral blood cysts (mean age, 48 ± 18 years), 9 with a tricuspid valve cyst (mean age, 67 ± 15 years), 3 with a blood cyst on the pulmonary valve (age 31, 43 and 44 years) and 1 aortic valve cyst in a 22-year-old man. Most patients were asymptomatic while stroke, syncope or myocardial infarction occurred in 6 patients with a mitral valve cyst. Blood cysts were removed surgically in 70% of patients with a mitral cyst, in 55% with a tricuspid cyst and in all those with a pulmonary or aortic cyst. At histology the cyst wall was composed mainly by fibrous tissue and with the inner surface lined with typical endothelium. Conclusions: Blood cysts of cardiac valves are rare in adults but may cause life-threatening complications particularly when located on the mitral valve. For such reason surgical removal appears advisable, with low-risk procedures. Widespread use of multimodality imaging techniques will most likely increase the number of valvular blood cysts diagnosed also in adults.

INTRODUCTION

Benign cardiac neoplasms are substantially rare and among these intracardiac cysts filled with blood are even more uncommon; indeed, they have not even been mentioned in a recent review of cardiac tumors reported by Tyebally et al.¹. However, mainly owing to advancement in diagnostic techniques, with a widespread use of and accessibility to multimodality imaging, an increase in the diagnosis of cardiac or pericardial masses has been observed recently and is expected to further grow in the future¹. Intracardiac blood cysts (BCs) are commonly found in newborns and infants being rare after the first year of life, they mainly involve the cardiac valves and generally disappear during growth². In adults they may occasionally be diagnosed, are usually located on all cardiac valves and chambers, the mitral valve (MV) being the most frequently involved; despite absence of histological malignancy BCs of the heart valves have been reported to cause life-threatening complications with even severe sequelae.

There is still uncertainty on the origin of BCs and controversy on whether medical treatment with continuous clinical and echocardiographic patient follow-up should be preferred to immediate surgical removal. The present review, analysing the published cases, was undertaken with the aim of assessing the incidence,

evaluating the clinical presentations and discussing the treatment of BCs involving the cardiac valves in adults.

METHODS

We have performed a search of the English literature, through PubMed and Medline, to identify all cases of BCs involving the MV, MV apparatus and tricuspid, aortic and pulmonary valves that have been so far reported. Articles in textbooks and meeting abstracts were excluded, as well as cases included in general reviews of cardiac tumors or clinical reports, when detailed clinical and pathological information on BCs, when present, was lacking. Cases of valvular cysts but with uncertain histological diagnosis were excluded. The reference sections of pertinent articles were also evaluated as well as personal files and the archives of journals available on the CTSNet website. All reported cases observed in the pediatric population were excluded, considering only those occurring in patients ≥ 18 years of age.

Main terms used in the literature search, alone or in combination, were: intracardiac cyst, valvular cyst, blood cyst, blood-filled cyst, pedunculated cysts, mitral valve, mitral valve apparatus, mitral valve leaflet, tricuspid valve (TV), papillary muscle, chordae tendineae, aortic valve (AV), pulmonary valve (PV), semilunar valve, valve cusps, atrio-ventricular valve, semilunar valve.

Approval by the Ethical Committees was not required for this kind of study, as well as informed patient consent, provided that all relative data were treated anonymously.

RESULTS

Blood cysts of the mitral valve

Our Literature search has documented a total of 50 articles considered eligible for analysis and reporting a total of 54 patients; 36 (72%) are reports of a single case³⁻³⁸, 12 (24%) images in cardiovascular disease³⁹⁻⁵⁰, 1 a case series reporting 5 patients (2%)⁵¹ and 1 a letter to the Editor (2%)⁵², all published from 1960 to 2021 (Table 1 and 2).

Patient characteristics : There were 29 males (55%) and 25 females (45%) with an age ranging from 18 to 87 years (mean, 48 ± 18 years). The majority of patients was asymptomatic and the cyst was disclosed at routine controls for other pathologies. Symptoms related to the presence of MV BCs, in the absence of any other intracardiac abnormality, included predominantly palpitations, chest discomfort and exertional dyspnea. In 3 patients BCs caused severe left ventricular outflow tract (LVOT) obstruction^{5,11,50}, 2 patients presented with hemiparesis^{4,43}, 2 with syncopal episodes^{32,50}, 2 had a stroke^{15,18}, 2 a myocardial infarction^{9,21} and in one the BC caused severe MV regurgitation²³. Except for the first 2 BCs cases described, diagnosed either by cineangiography³ or M-mode echocardiogram⁵, in all other cases transthoracic 2-D echo was used for evidencing the presence of a MV mass (Fig. 1). The diagnosis was confirmed by supplemental transesophageal echo in many cases and more recently by multimodality imaging, including angio-computed tomography, nuclear magnetic resonance, contrast real-time echocardiography and 3-D echo^{23,34,36,38,44,45,49,51}.

Surgical data : Cyst removal was performed in 37 patients (70%) while in 16 (30%) medical treatment and serial follow-up was recommended; in one patient aortic valve and ascending aorta replacement were performed leaving in place a small BC on the MV²⁰. Surgery was generally performed through a standard median sternotomy while in 3 cases a minimally invasive, endoscopic technique was used^{27,36,45}. Two patients had previous open heart operations and required repeat sternotomies^{22,42}, while one had a previous liver transplantation¹⁰. The BC was approached through a trans-aortic incision in 7 cases, through the left atrium in 6, trans-septal in 4, while in 20 cases it was not specified. Beside cyst removal, 27 patients (73%) had associated procedures: mitral valve replacement (MVR) in 12, mitral valve repair (MVR) in 9, coronary artery bypass grafting in 3 and combined MVR and aortic valve replacement in 1. In addition, in 2 patients, with associated *cor triatriatum*, resection of the intraatrial band was performed^{14,24}. No operative deaths are reported.

Blood cysts of the tricuspid valve

There are 9 articles on BCs of the TV, 6 case reports^{53,55-60}, 2 images in cardiovascular disease^{54,58} and 1 letter to the Editor⁶¹, published from 1991 to 2021 (Table 3).

Patient characteristics : Of the 9 patients 5 were females and 4 males with an age ranging from 35 to 88 years (mean, 67±15 years). Symptoms at presentation were generally not specific except for few cases with moderate-severe tricuspid regurgitation, with congestive heart failure or symptoms related to other concomitant cardiac pathology. One patient had an associated muscular ventricular septal defect without hemodynamic significance⁵⁶.

Surgical data : Cyst removal was performed in 6 cases (55%) while in the remaining 3, surgery was considered not indicated. All patients were operated through a median sternotomy using a right atriotomy for cusp excision; in one case even a combined left atrial-transseptal approach is reported. In 4 patients BCs removal was associated with various techniques of TV repair; in one patient myocardial revascularization was also performed.

Blood cysts of the pulmonary valve

Patient characteristics : Our search yielded 3 papers on BCs of the PV, 2 case reports^{62,63} and 1 image in cardiovascular disease⁶⁴ (Table 4). All were females, 31, 43 and 44 years of age. Auscultation of a cardiac murmur, associated to fatigue and dyspnea, prompted further evaluation with angiographic or echocardiographic disclosure of the BC. In 2 cases the pulmonary trunk had an aneurysmal dilatation^{62,64} and in one the PV was severely dysplastic⁶⁴.

Surgical data : Cyst removal was performed in all cases, by a transpulmonary approach. In one patient PV valvuloplasty was required associated to reduction of the pulmonary trunk size⁶⁴.

Blood cysts of the aortic valve

We found only a case of a BC, on an images in cardiovascular medicine paper, found on a bicuspid aortic valve in a 22-year-old male with aortic stenosis. The cyst had a maximum diameter of 21mm with a broad base of implant; it was excised during AVR⁶⁵

Blood cyst pathology

BCs of the cardiac valves are usually described as round or oval-shaped masses, either bluish or yellowish in colour, at times pedunculated and of variable sizes.

Mean maximum size of MV BCs was 20±8 mm, ranging from 3 to 46 mm; in 4 cases the cyst was considered as 'giant' despite a large variability of sizes^{7,9,26,32}. The most frequent location of the cysts was the anterior mitral valve leaflet (AML) occurring in 33 cases, followed by the anterior papillary muscle in 8, the posterior mitral leaflet (PML) in 5 and the posterior papillary muscle in 4. The cyst was described to be attached to the chordae tendinae in one, to the mitral annulus in one and to an unspecified papillary muscle in one; in one patient with 2 BCs both AML and PML were involved¹⁴. In 3 patients 2 BC were found^{6,14,30}. Some BCs have been described as multilobulated^{9,12,17,50}.

The maximum size of TV BCs ranged from 18 to 33 mm (mean, 24±5 mm); in 3 cases the size of the BC was not indicated^{54,58,61}. BCs were located on the septal TV leaflet in 4, anterior TV leaflet in 2 and the posterior TV leaflet in 1 case; in one patient location of the BC was not reported⁵⁸.

The maximum size of the 3 PV BCs was 8, 30 and 40 mm, respectively; according to the authors' description BCs were located on the posterior PV cusp in 1 case and on the right PV cusp in 1; in 1 case the location is not clearly indicated⁶⁴.

Histology of the excised tissue has not been reported in all the surgical cases. When available histologic data, regardless of BC location, have been substantially uniform in describing the BC wall as composed mainly by fibrous tissue of various thickness (Fig. 2A), at times with a myxoid stroma. In many cases the inner BC wall was lined with typical endothelium while smooth calcific spots were occasionally seen^{19,49}. The fibrous

nature of the BC wall is also confirmed by immunohistochemistry (Fig. 2B). Interestingly, no ultrastructural images have been reported in BCs excised from cardiac valves of human beings.

DISCUSSION

The first description of an intracardiac BC was reported by Elsässer as late as 1844⁶⁶, while Houser and colleagues are credited with the first use of echocardiography to identify such peculiar lesions⁴. In 1968 the first surgical removal of a MV BC was reported by Leatherman et al.³; as described by the authors ‘*The cyst was attached to the septal leaflet at the point where the leaflet was joined by chordae tendineae. During excision the cyst was opened and blood drained out. Extracorporeal circulation lasted 18 minutes, and recovery was uneventful*’³.

Intracardiac BCs are usually incidental necropsy findings or are diagnosed clinically predominantly in children, being extremely rare after the first year of life and particularly in adults^{1,2}. They are considered benign tumors, nevertheless, while benign from the histologic point of view, they have been associated to relevant complications such as LVOT obstruction and coronary or systemic embolization with consequent severe sequelae such as stroke or myocardial infarction^{4,5,9,11,15,18,20,48}. Transthoracic or transesophageal 2D echo are usually considered adequate to detect intracardiac masses even of small dimensions and the presence of an echo-free space within the mass may usually rise the suspicion of the presence of a blood cyst; others have considered formation of microbubbles inside the cyst, when using contrast real-time echocardiography, as pathognomonic of the presence of a BC^{12,17}. However, sometimes solid cardiac tumors, such as myxomas, may present with similar echocardiographic features therefore being difficult to be differentiated from a benign BC^{7,8}. Furthermore, it must be underlined that other intracardiac masses must be ruled out such as fibroelastomas or infectious vegetations which also frequently involve the cardiac valves and, therefore, the diagnosis of a BC is not always straightforward and needs histology to be confirmed. The potential role of magnetic resonance imaging has also been stressed in the diagnostic assessment of such patients particularly because of its specificity in ruling out signs of myocardial infiltration and therefore to exclude the presence of a malignant lesion²⁶.

From this review it appears that management of patients with a blood cyst is still controversial²¹. Many advocate surgical excision while according to others serial clinical and echocardiographic surveillance can be adequate especially in the presence of masses of small size to monitor the rate and degree of growth¹⁵. Indeed, in 30% of the patients analysed surgery was not considered either for BC small size, clinical stability, patient refusal or advanced age. However, due to the potential for embolization of even small cardiac masses, including BCs, we believe that surgical removal should always be indicated not only to prevent the risk of complications but also to determine or confirm the exact diagnosis.

The origin of BCs of the cardiac valves is still unknown. The first hypotheses were formulated in the early 1900’ by Bayne-Jones, studying the blood vessels of the heart valves⁶⁷. Large studies on hearts of dogs and mainly cows and calves obtained from slaughter houses, have demonstrated the presence of valvular BCs in almost 20% of hearts; the results of histological and ultrastructural evaluations support the hypothesis that BCs most likely derive from dilatation of the thin-walled valvular arteries due to the mechanical stresses induced by the pressure gradient when the atrio-ventricular valves are closed with consequent cyst formation^{68,69}. This theory, however, does not explain the occurrence of BCs in low pressure structures such as the pulmonary valve⁵⁵. Furthermore, it is not clear whether what observed in animal studies can be also be completely applied to human beings. According to Tsutsui et al., BCs could derive by blood trapped in valvular crevices or microscopic invaginations during development¹³; this might explain the finding of BCs in children but not BCs in adults with previously normal echocardiograms. Other consider BCs to derive from valvular hematomas²⁴, or being secondary to endocardial inflammation; accordingly, the few cases observed following previous open heart surgery might indicate an additional risk factor of cardiac surgery in the development of BCs, although this most likely is just an occasional association²².

This review has some limitations. Since we have excluded from our analysis cases possibly contained in specific textbooks or pathological reviews and those with an uncertain diagnosis, the number of MV BCs

may be underestimated. Even if the American Academy of Pediatrics has identified the upper age limit as 21 years for pediatric patients⁷⁰, we arbitrarily considered 18 years as the lowest age for this review; elevating this limit would have further reduced the number of recognizable cases. Although some of the data which could be obtained from single cases were not complete, this review provides enough evidences to assess the clinical presentation, diagnostic modalities and management of patients with a cardiac valve BC found in adult patients.

In conclusion, BCs are rarely found in patients [?]18 years of age and predominantly affect the MV. Although in most cases they represent an incidental finding, sometimes they are heralded by serious acute complications such as syncope and stroke. For such reason surgical removal, which is a low-risk procedure and can be performed with minimally invasive techniques, should be advisable for BCs of the MV, even if of small size; on the other hand, BCs in the right heart may be treated conservatively unless they cause TV regurgitation or obstruction to blood flow. The origin of valvular BCs is still quite uncertain but since their histological benign nature is well known further studies on pathogenesis would have only a speculative interest. It is likely that the current widespread use of multimodality imaging will increase the number of intracardiac BC detected and hopefully the present review will help to improve management of such patients.

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LEGENDS

Fig. 1: A) Transthoracic 2-D Echocardiographic four chamber view, in a 61-year-old asymptomatic man showing the intracardiac mass (*asterisk*) attached to the anterior mitral leaflet (AML) and B) moderate mitral regurgitation.

LA= Left atrium; LV= Left ventricle.

Fig. 2: A) Histologic section of the blood cyst wall showing the presence of fibrous tissue (Hematoxylin-eosin stain, original magnification x4). B) Immunostaining for the endothelial antigen CD31 (*brown colour*) highlighting the endothelial layer on the luminal surface (Immunoperoxidase, hematoxylin counterstaining, original magnification x 4).

Table 1: Characteristics of case reports and case series of blood cysts of the mitral valve apparatus.

Author, Ref. #	Year	Age, sex	Location	Max. dimension, mm	Surgical approach	Operation
Leatherman et al ³	1968	39,F	AML	30	Transaortic	Cyst removal
Hauser et al ⁴	1983	27,M	APM	25	LA incision	Cyst removal, MVR
Arnold et al ⁵	1990	46,M	AML	32	Transaortic	Cyst removal, MVR
Xie et al ⁶	1992	41,F	PML	13, 3*	Transseptal	Cysts removal
Ohmoto et al ⁷	1993	57,M	APM	40	Transaortic	Cyst removal
Pelikan et al ⁸	1999	50,M	AML	22	No surgery	-
Sharma et al ⁹	2000	68,M	APM	20	Transaortic	Cyst removal, CABG
Kuvin J et al ¹⁰	2004	45,F	AML	25	LA incision	Cyst removal
Minnecci et al ¹¹	2004	44,F	AML	20	Transaortic	Cyst removal, MVR
López-Pardo et al ¹²	4008	34,M	AML	22	No surgery	-

Author, Ref. #	Year	Age, sex	Location	Max. dimension, mm	Surgical approach	Operation
Tsutsui et al ¹³	2008	47,F	AML	15	No surgery	-
Denker et al ¹⁴	2009	65,F	AML, PML	10, 10*	NA	Cysts removal, MVr, LA band resection**
Lodha et al ¹⁵	2009	74,F	AML	NA	No surgery	.
Park et al ¹⁶	2009	22,M	APM	21	NA	Cyst removal, MVr
Migliore et al ¹⁷	2010	18,F	AML	22	No surgery	-
Khan et al ¹⁸	2012	87,F	AML	17	No surgery	-
Park et al ¹⁹	2012	47,M	APM	18	NA	Cyst removal, MVr
Bhatt et al ²⁰	2013	45,F	PML	NA	-	No removal
Donndorf et al ²¹	2013	55,M	PM (n.s.)	20	Transaortic	Cyst removal, CABG
Ansari et al ²²	2015	55,M	AML	26	LA incision	Cyst removal, CABG
Halim et al ²³	2015	23,M	AML	20	Transseptals	Cyst removal, MVr
Madhavan et al ²⁴	2015	70,F	AML	16	NA	Cyst removal, MVR
Özmen et al ²⁵	2015	19,F	AML	17	NA	Cysts removal, RA band resection**
Yilmaz et al ²⁶	2015	63,M	AML	17	No surgery	-
Ahmad et al ²⁷	2016	25,M	PPM	20	NA	Cyst removal, MVr
Okamoto et al ²⁸	2016	41,F	PPM	29	LA incision	Cyst removal, MVR
Akutsu et al ²⁹	2017	57,M	AML	15	Transseptal	Cyst removal, MVr

Author, Ref. #	Year	Age, sex	Location	Max. dimension, mm	Surgical approach	Operation
Pavsic et al ³⁰	2017	44,F	AML	23	NA	Cyst removal, MVR
Bagheri et al ³¹	2018	62,M	PPM	21, 19*	NA	Cysts removal, MVR
Ludhwani et al ³²	2019	47,F	MVA	15	No surgery	-
Ma et al ³³	2019	32,M	AML	46	NA	Cyst removal
Cerik et al ³⁴	2020	42,F	AML	17	No surgery	-
Ramirez-Mesias et al ³⁵	2020	57,F	AML	10	No surgery	-
Wang et al ⁵¹	2020	30,M 45,M 57,F 58,M 57,M	AML AML AML AML MV chordae	17 25 11 19 12	NA NA NA NA No surgery	Cysts removal Cyst removal, MVR Cyst removal, MVR Cyst removal -
Beale et al ³⁶	2021	62,F	AML	20	No surgery	-
Ionac et al ³⁷	2021	39,M	AML	40	NA	Cyst removal, MVr
Zhang et al ³⁸	2021	38,M	APM	25	LA incision	Cyst removal

M= Male; F= Female; NA= Not available; AML= Anterior mitral leaflet; MVr= Mitral valve repair; PML= Posterior mitral leaflet; MVR= Mitral valve replacement; CABG= Coronary artery bypass grafting; APM= Anterior papillary muscle; PPM= Posterior papillary muscle.; LA= Left atrium; RA= Right atrium; MV= Mitral valve; PM= Papillary muscle; n.s.=not specified; MVA= Mitral valve annulus.

* These patients had 2 cysts; ** These patient had a *cor triatriatum* sinister and dexter, respectively;[§] This patient had a re sternotomy due to a previous closure of an atrial septal defect.

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Table 2: Characteristics of cases reported as images or letter to the Editor of blood cysts of the mitral valve apparatus.

Author, Ref. #	Year	Age, sex	Location	Max. dimension, mm	Surgical approach	Operation
Kurtoğlu et al ³⁹	2005	35,M	AML	21	No surgery	-
Yamamoto et al ⁴⁰	2005	25,F	AML	25	NA	Cyst removal,
Akinci and Celikyurt ⁴¹	2010	64,M	AML	18	No surgery	PTCA
Combaret et al ⁴²	2012	66,M	AML	16	NA	Cysts removal
Grimaldi et al ⁴³	2012	63,F	AML	15	No surgery	-
Kuhn et al ⁴⁴	2012	28,F	PML	22	Transseptal*	Cyst removal
Paluszkiwicz et al ⁴⁵	2013	80,F	APM	25	NA	Cyst removal
Sun et al ⁴⁶	2013	25,M	AML	NA	No surgery	-

Author, Ref. #	Year	Age, sex	Location	Max. dimension, mm	Surgical approach	Operation
Bonenfant et al ⁴⁷	2014	25,F	AML	19	NA	Cyst removal
Dubey et al ⁴⁸	2017	22,F	PML	NA	NA	Cyst removal,
Bezjak et al ⁴⁹	2019	63,M	AML	24	LA incision	Cyst removal,
Xiao et al ⁵⁰	2019	32,M	APM	46	NA	Cyst removal,
Romano et al ⁵²	2009	71,M	PPM	21	NA	Cyst removal,

M= Male; F= Female; NA= Not available; AML= Anterior mitral leaflet; MVr= Mitral valve repair;

PML= Posterior mitral leaflet; MVR= Mitral valve replacement; PTCA= Percutaneous transluminal coronary angioplasty; AVR= Aortic valve replacement; APM= Anterior papillary muscle; PPM= Posterior papillary muscle.

* This patient had a second re sternotomy due to a previous repair of corrected transposition of the great arteries, ventricular septal defect and pulmonary stenosis with a Rastelli-type of procedure followed by pulmonary homograft implantation.

Table 3: Characteristics of reported cases of blood cysts of the tricuspid valve.

Author, Ref. #	Year	Age, sex	Location	Max. dimension, mm	Surgical approach	Operation
Paşaoğlu et al ⁵³	1991	35,M	STL	30	RV	Cyst removal
Timperley et al ⁵⁴	2004	80,F	STL	NA	RA*	Cysts removal
Michelena et al ⁵⁵	2007	55,M	ATL	30	RA	Cysts removal, TVr
Agac et al ⁵⁶	2009	72,F	STL	20	No surgery	-
Grapsa et al ⁵⁷	2011	88,F	STL	25	No surgery	-
Butler et al ⁵⁸	2015	66,M	NA	NA	RA	Cysts removal, TVr
Kalçık et al ⁵⁹	2015	65,M	PTL	18	LA-Transseptal	Cysts removal, TVr
Aydin et al ⁶⁰	2019	68,F	STL	23	No surgery	-
Taylan et al ⁶¹	2021	74,F	ATL	NA	RA	Cyst removal, TVr

M= Male; F= Female; NA= Not available; STL= Septal tricuspid leaflet; ATL= Anterior tricuspid leaflet;

RV= Right ventriculotomy; RA= Right atriotomy; TVr= Tricuspid valve repair; PTL= Posterior tricuspid leaflet; CABG= Coronary artery bypass grafting; NA= Not available.

* Unless specifically indicated, cyst removal was considered performed though a right atriotomy.

Table 4: Characteristics of reported cases of blood cysts of the pulmonary valve.

Author, Ref. #	Year	Age, sex	Location	Max. dimension, mm	Surgical approach	Operation
Liese et al ⁶²	1963	31,F	PPC	40	Transpulmonary	Cyst removal
Minato et al ⁶³	1997	43,F	RPC	8	Transpulmonary	Cyst removal
Zhang et al ⁶⁴	2021	44,F	NA	30	Transpulmonary	Cyst removal, PV r

F= Female; PPC= Posterior pulmonary cusp; RPC= Right pulmonary cusp; PV= Pulmonary valve:

PA= Pulmonary artery.

* This patient had a dysplastic pulmonary valve.



