

# Successful surgical management of ventricular fibromas in children

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**Objectives:** A uniform management strategy has not been established for the treatment of large ventricular fibromas identified in children. The goal of the present study was to review our experience with surgical resection of this uncommon tumor.

**Methods:** We performed a retrospective review of all patients with a diagnosis of ventricular fibromas who had undergone surgical intervention at our institution from July 1990 to June 2013. The clinical records and pathology reports were reviewed and available follow-up data obtained.

**Results:** A total of 20 patients had a ventricular fibroma. The median age at presentation was 4 years (range, 5 months to 12 years). Of these, 18 (89%) presented with ventricular tachycardia (n = 15) or frequent ectopy (n = 3), and 1 (6%) with outflow tract obstruction; 1 (6%) was asymptomatic and diagnosed during routine evaluation for a murmur. One patient had undergone previous ablation, and one had had an automated internal cardiac defibrillator implanted for ventricular tachycardia control. Before tumor resection at our institution, 1 patient had been listed for transplantation at an outside institution and 1 had undergone previous extracardiac Fontan without tumor resection. All patients underwent successful tumor excision with no recurrence of ventricular arrhythmia during a median follow-up period of 3.3 years (range, 1 month to 14.7 years). One patient underwent planned staged tumor resection. No patient died.

**Conclusions:** Primary ventricular fibromas, even when very large, can be managed by surgical resection, with excellent short- and intermediate-term ventricular function and relief of arrhythmic events. (*J Thorac Cardiovasc Surg* 2014;148:2602-8)

Benign cardiac tumors represent 75% of all cardiac tumors, which have an incidence of 0.001% to 0.3% in autopsy series. Cardiac fibromas represent the second most common benign pediatric tumor.<sup>1-3</sup> Autopsy studies of fibroma were reported as early as 1855, and the first case reports were published in 1949.<sup>4</sup> Since then, these tumors have been described in numerous case reports.<sup>5,6</sup> Although major strides have been made in establishing the diagnosis, especially with noninvasive imaging,<sup>7</sup> management strategies, in particular, surgical strategies, have not been well-defined.<sup>8-23</sup>

The goal of the present study was to review our institutional experience with surgical management of ventricular fibromas, with special attention to the surgical techniques used for successful resection and amelioration of the potentially life-threatening arrhythmias. We chose to focus on the outcomes of patients who had undergone surgical management of symptomatic ventricular fibroma,

because reports have varied on the ideal management of these tumors and have included resection, transplantation, and single ventricle palliation.

## METHODS

A retrospective review of all patients with a tissue diagnosis of ventricular fibroma who had undergone tumor resection at our institution from July 1990 to June 2013 was performed. The institutional review board approved the conduct of our review. We queried our cardiac surgical, cardiology, echocardiography, and pathology databases during the study period for all patients who presented to our institution with a diagnosis of fibroma. The clinical records were reviewed for clinical history and operative data. The reports of all pre- and postoperative imaging studies, including echocardiograms; magnetic resonance imaging, cardiac catheterization, and electrophysiologic studies; electrocardiograms; and Holter monitor studies were reviewed. The pathology reports were examined for confirmation of the diagnosis. All operative reports were reviewed to document the surgical technique at resection. Special attention was given to the tumor location and completeness of resection.

## RESULTS

From 1990 to 2013, 153 patients were identified with cardiac tumors: 92 rhabdomyoma, 26 fibroma, 10 myxoma, 5 vascular, 3 teratoma, 3 lipoma, and 14 other tumors. In this group of 153 patients, 53 underwent cardiac tumor resection, with resection of ventricular fibromas in 20 patients. All patients who were referred to our center for symptoms underwent resection; none were deemed inoperable or considered for palliative procedures, such as single ventricle palliation, and none were evaluated for

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**Abbreviation and Acronym**  
 ECMO = extracorporeal membrane oxygen

transplantation. Of these 20 tumors, 18 were located in the left ventricle and 2 in the right. The distribution between males and females was equal (10 each). The median age at resection was 4.2 years (range, 5 months to 12 years). The median follow-up duration was 3.3 years (range, 0.1-14.7 years). No patient died. One patient who had had 2 separate large tumors required staged resection, with removal of the anteroseptal tumor first, followed by removal of the second tumor arising in close proximity to the atrioventricular groove, in a subsequent procedure. Seven patients required additional intracardiac work (atrioventricular valve repair in 5, patent foramen ovale closure in 1, and Fontan conversion in 1). Of the 20 patients, 18 (90%) presented with some form of ventricular arrhythmia, ranging from frequent premature ventricular beats to sustained or nonsustained ventricular tachycardia; with 1 patient experiencing a ventricular fibrillatory arrest. One patient was referred after undergoing staged single ventricle management, including Stansel connection at birth and, later, an extracardiac Fontan. One patient had been deemed inoperable and listed for transplantation at an outside

institution, before successful tumor resection at our institution. There was no recurrence of arrhythmia postoperatively in any of the 20 patients during early and late follow-up. The patient and tumor characteristics are listed in Table 1, the imaging characteristics in Table 2, and the individual patient data in Table 3. A graphic representation of the tumor location is presented in Figure 1. Figure 2 shows the magnetic resonance imaging, intraoperative, and histologic findings from a representative case.

**Surgical Technique**

All procedures were performed with cardiopulmonary bypass and moderate hypothermia (28°C in 11, 25°C in 5). More recently, 7 patients underwent this procedure using a lower hemisternotomy with excellent tumor visualization. All but 2 patients had undergone bicaval cannulation. Early in our experience, cardioplegic arrest was used in 3 patients. In the more recent era (since 2004), resection has been performed with a beating heart in 5. In the other 12 patients, tumor resection was performed with the patient under fibrillatory arrest (induced in 6 and spontaneous in 6), because this was believed to allow better delineation of the epicardial coronary arteries that needed to be preserved. Six patients had undergone tumor resection with a beating or fibrillating heart and then underwent cardioplegic arrest

**TABLE 1. Summary of patient and procedure characteristics (n = 20)**

Characteristic	Value
<b>Preoperative</b>	
Male gender	10 (50%)
Median age (y)	4.21 (0.44-12.45)
Weight (kg)	16.9 (6.9-55.9)
<b>Preoperative findings</b>	
Asymptomatic	1
Ventricular arrhythmias	18
RVOTO	1
<b>Intraoperative</b>	
Median on-pump time (min, n = 20)	112 (62-363)
Median fibrillatory time (n = 8)	52 (5-233)
Median crossclamp time (n = 10)	70 (8-98)
<b>Associated procedure (6/20 patients [30%])</b>	
Biventricular conversion from Fontan	1
Mitral valve repair	4
Tricuspid valve repair	1
PFO closure	3
<b>Postoperative</b>	
ECMO	2
Median length of stay (d)	7 (4-33)
Median follow-up (y) (n = 17/20*)	3.3 (1 mo to 14.7 y)
Death	0
Reoperation	0
Ventricular arrhythmia	0

Data presented as median (range) or n (%). RVOTO, Right ventricular outflow tract obstruction; PFO, patent foramen ovale; ECMO, extracorporeal membrane oxygen. \*The 3 patients lost to follow-up were all from outside the United States.

**TABLE 2. Preoperative and postoperative findings**

Variable	Preoperative findings (n = 20)	Postoperative findings (n = 14)
<b>LVEF echocardiogram (n)</b>		
>55%	19	11
40%-55%	1	3
<40%	0	0
<b>LVEF MRI</b>		
Mean ± SD	61 ± 10	NA
Median	64	
Range	35-72	
<b>LVEDVi MRI</b>		
Mean ± SD	78 ± 18	NA
Median	81	
Range	38-115	
<b>Tumor length (cm)</b>		
Mean ± SD	5.4 ± 1.3	NA
Median	5	
Range	3-8.6	
<b>Associated findings (n)</b>		
Residual AVVR*	NA	4
Residual tumor†		8
RVOT aneurysm		1
Arrhythmia	18	0

LVEF, Left ventricular ejection fraction; MRI, magnetic resonance imaging; SD, standard deviation; NA, not applicable; LVEDVi, left ventricular end-diastolic volume indexed; AVVR, atrioventricular valve regurgitation; RVOT, right ventricular outflow tract. \*Unchanged or improved from preoperatively. †Thin rim adjacent to vital structures.

**TABLE 3. Detailed patient characteristics of ventricular fibromas (presenting symptoms, tumor location, follow-up data)**

Pt. no.	Age	Presentation	Preoperative workup	Tumor location	Preoperative arrhythmia
1	7.5 mo	Murmur RVOTO	Echo MRI	RV free wall to left of RVOT, with RVOTO	None
2	7 mo	URI symptoms, FTT	Echo, MRI	Anterolateral LV free wall from base to ALPM and extending over proximal LCA	Holter 132 VPB
3	6 y	Palpitation	Echo, MRI	PL LV free wall from base to apex	Holter 438 PVCs, 74 runs of VT
4	6 y	Ectopy during T&A	Echo, MRI	Inferoseptal and apical wall of LV	Vent ectopy
5	3 y	Murmur	Echo, EPS, Cath, MRI	Intramyocardial LV mass involving high septum and anterolateral wall	Holter-nonsustained VT, PVCs, couplets, bigeminy
6	10.5 y	CXR for URI symptoms	Echo, Cath	LV apical anterolateral free wall and anterior septum	ETT isolated, multifocal and slow ventricular couplets, ventricular triplets, and one 4-beat run of slow VT
7	12.5 y	Tachycardia during a febrile illness	Echo, MRI EPS, Cath	Anterolateral free wall	Sustained monomorphic VT
8	4.5 y	CXR for URI	Echo, Cath	LV free wall from level of ALPM to just above LV apex	Holter- occasional PVC, 2 couplets, 1 triplet, one 6 beat run of accelerated idioventricular rhythm
9	11 y	ER admission with SOB	Echo	Apical LV mass, extension into ALPM	Recurrent VT
10	6 y	Wide complex tachycardia hemiparesis, factor V Leiden mutation	Echo, MRI	Lateral aspect of LV free wall, possible involvement of ALPM	Sustained VT
11	4 y	Vfib arrest at 2 mo	Echo, MRI	Tumor involving inferior and inferolateral wall of LV, closely associated with PMPM	Vfib arrest at 2 mo, subsequent runs of monomorphic VT on Holter despite antiarrhythmic agents
12	4 y	Wide complex tachycardia	Echo, EPS, Cath	Tumor along inferolateral wall of LV	Wide complex tachycardia, EPS 1, spontaneous self-resolving VT, inducible VT × 2 foci requiring cardioversion
13	8 mo	Murmur	Echo, Cath, MRI	IVS from base to apex, abutting PMPM and commissure	No preoperative arrhythmia, referred to surgery for increasing tumor size
14	5 mo	Diagnosis on fetal Echo	Echo, Cath, EPS, Cath	Lateral wall of LV extending from base to apex	Monomorphic VT, inducible VT on EPS
15	4.5 y	Presyncope	Echo, MRI	Anterolateral and lateral wall from base to midway to apex	Presyncope nonsustained VT
16	2 y	Murmur	Echo, MRI, Cath	LV free wall extending from base to 2/3 of distance to apex, mild sub-AS	Non sustained VT on Holter, inducible poly- or monomorphic VT on EPS
17	10 mo	VT	Echo, MRI, CT	Inferior LV wall extending from base to apex, no OTO	VT
18	5.3 y	Ventricular arrhythmias	Echo, MRI	Anteroseptal wall extending to base of LV	Premature ventricular beats, unifocal
19	7.5 mo	VT	Echo, MRI, Cath	Anteroseptal, anterior and anterolateral segments, extending from base to nearly apex	VT
20	8.5 y	VT	Echo, MRI	Anterior free wall right ventricle	Asymptomatic VT on Holter

*Pt. no.*, Patient number; *RVOTO*, right ventricular outflow tract obstruction; *FU*, follow-up; *Echo*, echocardiogram; *MRI*, magnetic resonance imaging; *RV*, right ventricular/ventricle; *RVOT*, RV outflow tract; *URI*, upper respiratory infection; *FTT*, failure to thrive; *LV*, left ventricular/ventricle; *ALPM*, anterolateral papillary muscle; *LCA*, left coronary artery; *VPB*, ventricular premature beat; *PL*, posterolateral; *PVCs*, premature ventricular contractions; *VT*, ventricular tachycardia; *T&A*, tonsillectomy and adenoidectomy; *EPS*, electrophysiologic study; *Cath*, cardiac catheterization; *CXR*, chest radiograph; *ETT*, exercise test; *LAD*, left anterior descending (artery); *LCA*, left coronary artery; *EF*, ejection fraction; *ER*, emergency room; *AICD*, automatic implantable cardioverter defibrillator; *AV*, atrioventricular; *PFO*, patent foramen ovale; *Vfib*, ventricular fibrillation; *IVS*, interventricular septum; *PMPM*, posteromedial papillary muscle; *MV*, mitral valve; *AS*, aortic stenosis; *MR*, mitral regurgitation; *OTO*, outflow tract obstruction.

for inspection of the atrioventricular valve or subaortic region or for patent foramen ovale closure. The left heart was decompressed by a right superior pulmonary vein vent. Once cardiopulmonary bypass had been initiated,

careful examination of the tumor for its proximity to the major coronary arteries and atrioventricular groove was performed, and the ideal incision site for resection was determined. The epicardial incision was made, taking care



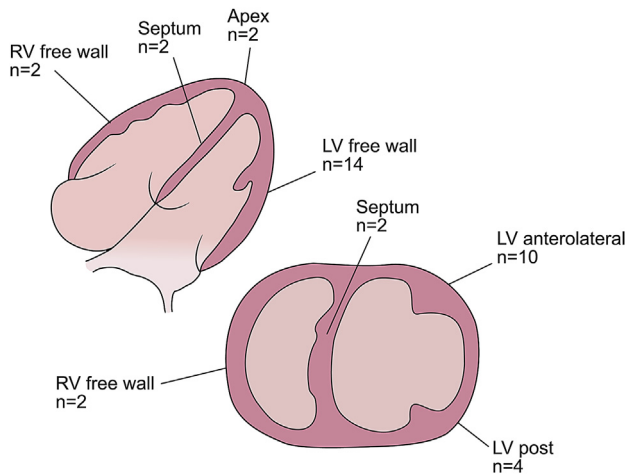
TABLE 3. Continued

Surgery	FU duration	Reintervention after discharge	Current status
Complete resection of RV mass	14.7 y	None	2.8 × 1.5 cm RVOT aneurysm, no arrhythmia
Near complete resection of LV fibroma	11.5 y	None	Mild global LV systolic dysfunction, no arrhythmias
Complete resection of LV tumor	1 mo	None	Lost to FU
Complete resection of LV apical tumor	1 mo	None	Lost to FU
Subtotal Excision of LV fibroma, (thin rim of tumor to protect epicardial coronaries and septum)	8.6 y	None	Normal LV function on echo, residual tumor on MRI in anterior basal septum, no arrhythmias
Resection of LV tumor, a thin rim of residual tissue beneath LAD, PFO closure	8.4 y	None	Apex and apical portion of LV thin and slightly echo-bright, mildly generous aortic annulus, no arrhythmia
Excision of LV tumor, with a thin rim of tumor beneath LCA and ventricular septum	8.9 y	None	Mildly dilated LV with minimally depressed function EF 49.7%, Holter: VPBs with no runs of VT
Complete excision of LV tumor	9 y	None	Good LV function, Holter single isolated PVC
Complete excision of tumor, and removal of AICD leads	6.9 y	None	Low normal LV function, EF 42.8%, no arrhythmia
Excision of tumor leaving a thin rim of tumor at AV groove and under diagonal branch, PFO closure	5.5 y	None	Residual hemiparesis, no significant change in residual tumor size, no arrhythmia
Complete tumor resection	4 y	None	Well, no arrhythmia
Total tumor excision	1 mo	None	Lost to FU
Enucleation of apical tumor, MV plasty, and sub-AS inspection	3.7 y	Planned staged resection LV, septal fibroma mitral valvuloplasty, sub-AS resection	Mild LV dysfunction, EF 42%, moderate MR unchanged from previously, no LVOTO, no residual tumor, no arrhythmia
Resection of tumor, leaving behind a thin rim of tumor at AV, MV plasty	2.5 y	None	Mild global LV dysfunction, improved from previous, large cavernous region near LV apex, mild MR, no LVOTO Holter: 3 PVC, 3 PAC
Complete tumor excision	3 y	None	Well, no arrhythmia, no recurrence
Major tumor debulking, ligation of coronary branch running into tumor	1.5 y	None	Well, no arrhythmia
Total excision of LV fibroma, MV repair	6 mo	None	Well, no arrhythmia
Excision of LV fibroma biventricular conversion from Fontan, ECMO	5 mo	None	Well, no arrhythmia
Excision of LV fibroma, mitral valvuloplasty, ECMO	4 mo	None	Well, no arrhythmia
Excision of RV fibroma, tricuspid valvuloplasty, PFO closure	2 mo	None	Well, no arrhythmia

to avoid the coronary arteries, at the point of maximal prominence of the tumor, and the incision was continued parallel to the major coronary arteries. The plane between the epicardium and tumor was developed and was often

well-delineated, because the tumor had a typical whitish fibrous texture. Every attempt was made to enucleate the entire tumor. Occasionally, transmural resection with entry into either ventricular cavity was required. A thin layer of



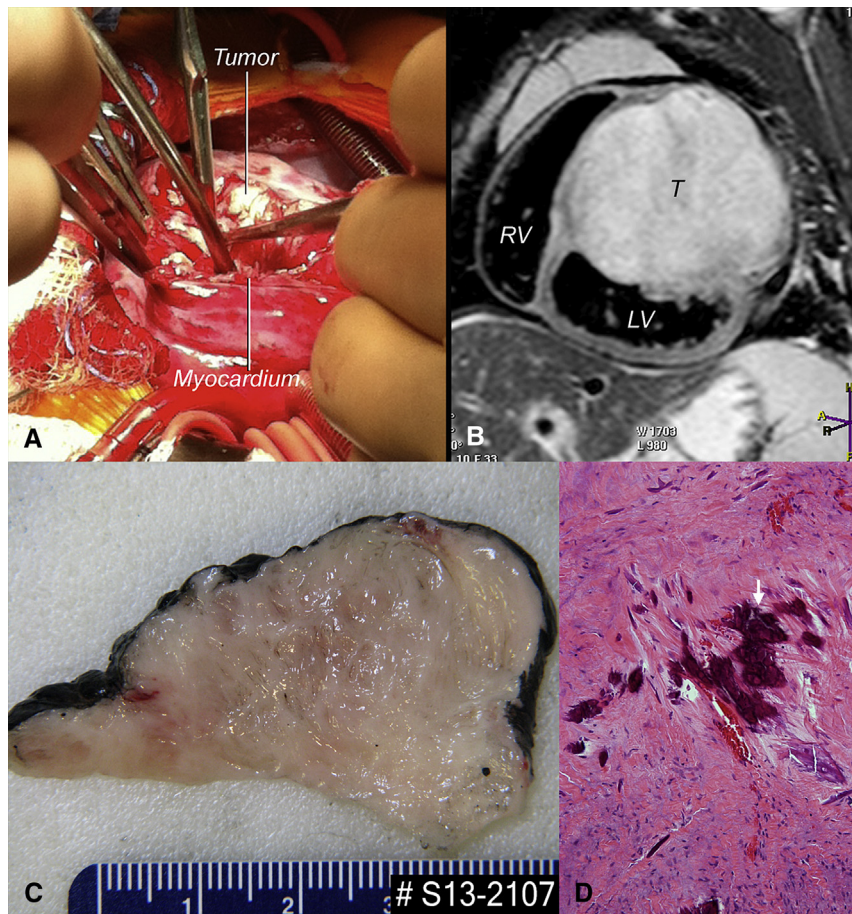


**FIGURE 1.** Location of fibromas. Most were in the left ventricular (LV) free wall. RV, Right ventricular.

residual tumor was left behind if it had extended close to the atrioventricular groove or close to a major epicardial

coronary artery. Once complete or near complete tumor resection had been confirmed by the presence of viable myocardial tissue in the tumor bed, the tumor bed was obliterated with interrupted pledgetted Prolene sutures in layers, taking care to avoid any kinking of the epicardial coronary arteries. In most cases, the epicardium was reapproximated over the obliterated tumor bed with fine Prolene sutures. If any concern for coronary compromise was present, the tumor bed was left partially open. Any intracardiac work that was required (eg, inspection and/or repair of the atrioventricular valve, inspection of the left ventricular outflow tract, closure of a patent foramen ovale) was then performed with the patient under cardioplegic or fibrillatory arrest. The left heart was de-aired, and the patients were weaned off cardiopulmonary bypass once they had been rewarmed. Atrial and ventricular wires were routinely placed, and all but 4 patients underwent primary chest closure.

Concomitant atrioventricular valve repair was performed in 5 patients (4 mitral valve and 1 tricuspid valve). This



**FIGURE 2.** Resection of a large left ventricular fibroma in a 7-month-old girl with ventricular tachycardia. A, Intraoperative photograph showing resection of the tumor along the plane of the normal myocardium. B, T<sub>1</sub>-weighted magnetic resonance image of the tumor in the ventricular short-axis showing the anatomic features of the mass. C, Gross pathologic specimen showing a somewhat inhomogeneous white-gray solid tumor mass. D, Photomicrograph showing dense collagen, focal necrosis, and calcifications (arrow). LV, Left ventricle; RV, right ventricle; T, tumor.

included release of tethering cords of the anterior mitral leaflet at the posteromedial commissure, reapproximation of the anterior mitral leaflet, and annuloplasty; partial closure of the anterolateral commissure, which was splayed open; anterolateral and posteromedial commissuroplasty for prolapse of the posterior mitral leaflet; release of the abnormal attachments at anterolateral commissure with partial closure of the commissure; and anteroseptal commissure closure and commissuroplasty to reduce tricuspid valve annulus. During follow-up, all patients had stable or an improved grade of atrioventricular valve regurgitation.

Only 3 patients experienced ventricular tachycardia with weaning off of cardiopulmonary bypass. One responded to intravenous lidocaine and a lidocaine infusion was continued for 72 hours postoperatively. The second required reinstatement of cardiopulmonary bypass and take down of the left internal mammary artery in preparation for coronary artery bypass grafting owing to concerns of ischemia in the circumflex coronary territory. However, this patient subsequently recovered normal function and was free of ectopy after a period of support with cardiopulmonary bypass and did not undergo coronary artery bypass graft. In both these patients, the chest was left open, with tourniquets in place; however, they did not require any additional intervention, and the chest was closed within the next 24 hours in the first patient and 72 hours in the second patient. The third patient was weaned off bypass onto extracorporeal membrane oxygen (ECMO) support for ventricular arrhythmias that had responded to lidocaine. This patient required ECMO for 3 days and was successfully decannulated.

In addition, the patient who had undergone tumor resection followed by biventricular conversion from Fontan circulation was electively supported with ECMO for left atrial hypertension, with a plan for continued postoperative mechanical support to allow for left ventricular remodeling. This patient continued with ECMO support for 5 days and required peritoneal dialysis for acute renal failure. The patient was successfully decannulated on postoperative day 5, with subsequent resolution of the renal dysfunction.

Most patients were extubated within 24 to 72 hours of surgery (except for the 2 patients requiring mechanical support; 1 remained ventilated for 14 days and 1 for 8 days). Only 1 patient had a period of junctional rhythm postoperatively; however, no ventricular arrhythmia was documented in the postoperative period in any patient. The findings of a postoperative ventricular stimulation study in 7 patients using their epicardial wires were all negative. Of the 20 patients, 14 (70%) had been placed on antiarrhythmic agents preoperatively and 11 (55%) were discharged with antiarrhythmic medication. During the follow-up period, 4 (24%) of the 17 patients with long-term follow-up data available continued with

antiarrhythmic medication as prophylactic therapy. None of the 17 patients had evidence of ventricular tachycardia or fibrillation on Holter monitoring during the follow-up period.

## DISCUSSION

Ventricular fibromas are rare primary cardiac tumors. In our series, we treated 20 patients from 1990 to 2013, with 19 of the 20 treated after 2001. The management strategy, in particular, the surgical approach has not been well established. The large tumor size in relationship to the ventricular chamber has often been considered a limiting factor for complete excision. In our experience, all but 1 of the tumors could be resected in their entirety, independent of the tumor size, tumor location, or patient age.

Several isolated case reports and small case series of complete or partial excision<sup>8-15</sup> have been published, and these have included techniques such as the Batista ventriculoplasty and Dacron patch sandwich for closure of the tumor bed after resection. The alternative management approaches to cardiac fibroma reported by others included functional single ventricle palliation,<sup>16</sup> single ventricle palliation as a bridge to transplantation,<sup>17</sup> partial excision with single ventricle palliation,<sup>18</sup> initial single ventricular palliation followed by resection,<sup>19</sup> staged resection,<sup>20</sup> and orthotopic heart transplantation as a primary surgical option for large fibromas, in particular, those involving the interventricular septum.<sup>21,22</sup>

The largest series to date on the management of cardiac fibroma was reported by Burke and colleagues,<sup>23</sup> who reported on 23 patients, including adults and children. Of these, 21 had ventricular fibromas; 13 (8 children and 5 adults) underwent resection (8 simple excisions, 4 incomplete excisions, and 5 complex resections, including patching of myocardial defects, valve replacement, or coronary artery bypass grafting). They reported 2 deaths in this group of 13. The group from the Mayo Clinic<sup>24</sup> reported their experience with surgical resection of ventricular fibromas from 1964 to 2002. That cohort included 12 children (median age, 3 years; range, 0.2-12.4) and 6 adults. Three patients had been referred to their center for cardiac transplantation. They had 1 intraoperative death. Similar to our experience, they reported no recurrent arrhythmias or tumor recurrence in the patients who survived surgery (median follow-up, 8.6 years; range, 0.4-33.4), but 2 did require subsequent mitral valve replacement.

Recent review at our institution<sup>25</sup> of a retrospective series of cardiac tumors and associated arrhythmias during a 42-year period, found that surgical excision was effective in the management of arrhythmias. This study included 13 of the patients in the current report.

The present series of 20 patients with cardiac fibromas in multiple locations has demonstrated that surgical excision

and, in most cases, complete enucleation, can be safely performed, with acceptable postoperative ventricular function and relief from potentially life threatening arrhythmia. The mid-term follow-up data have shown no recurrence of major arrhythmia or tumor. Late follow-up data from this group are necessary to determine the long-term efficacy of this approach, although the present length of follow-up would indicate that ventricular dysfunction would not appear to be a problem, despite the magnitude of the tumor at excision. Although single ventricle palliation or orthotopic transplantation could be considered as an alternative, all our patients were successfully treated with, predominantly, complete excision. In the 8 patients who underwent near total excision with a small rim of residual tumor, none have required reoperation to date, and none have developed recurrent ventricular arrhythmia.

### Study Limitations

The present study was a retrospective review; therefore, the follow-up data were not complete for all patients, with a median follow-up period of only 3.4 years. Three patients were lost to follow-up, because they had been referred to us from outside the United States. Despite extensive effort, we were unable to obtain any follow-up information. In addition, 4 patients from our cohort of 20 had undergone surgery within the previous 12 months (3 in the previous 6 months). If we excluded those lost to follow-up and those treated within the past 1 year, the median follow-up period would be 6.9 years (range, 2.5-11.5) for those treated since 2001.

Our review only included patients who had undergone surgical resection for symptomatic ventricular fibroma at our institution. Our study was not a comprehensive review of all ventricular tumors. In the same period, 6 additional patients had a diagnosis of fibroma determined by the magnetic resonance imaging characteristics. All these patients were asymptomatic, with no history of arrhythmia, and are followed conservatively.

### CONCLUSIONS

Large ventricular fibromas can be safely managed with primary total or near total excision, with relief of life-threatening arrhythmias and excellent clinical results.

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