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Surgical Experience With Cardiac Papillary Fibroelastoma Over a 15-Year Period

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Abstract

Background—Papillary fibroelastomas are rare, benign cardiac tumors. They are predominantly asymptomatic. However, they can lead to serious complications, namely thromboembolic events. Symptomatic lesions can be managed primarily with surgical excision and valvular preservation. Controversy exists as to the management of asymptomatic lesions.

Methods—All patients diagnosed with cardiac papillary fibroelastoma between 1996 and 2012 at a single institution were queried for clinical and pathologic characteristics.

Results—Twenty-three patients with 29 lesions were identified. Most lesions were solitary, less than 1.0 cm in diameter, and occurred in patients greater than 60 years of age. The most common presentation was thromboembolic complication. All were managed successfully with surgical excision. One patient developed a recurrence or metachronous lesion within 3 months of initial surgical intervention.

Conclusions—Papillary fibroelastomas are rare, benign, predominantly asymptomatic cardiac tumors that can cause potentially serious complications. The natural history and etiology of papillary fibroelastomas are largely unknown. Controversy exists over the management of asymptomatic lesions. However, there is consensus that symptomatic lesions should undergo surgical excision with valvular preservation when possible. A unique case of a possible papillary fibroelastoma recurrence is also described.

Cardiac papillary fibroelastomas (PFE) are benign cardiac neoplasms, with an incidence of 0.33% in autopsy series [1]. They are the third most common benign cardiac tumors and are the most common valvular tumors [2, 3]. Although rare and predominantly asymptomatic, they can be associated with serious complications, namely thromboembolic events.

This study was performed to review the surgical experience with papillary fibroelastoma at a single institution over a 15-year period. Twenty-three patients underwent surgical excision for PFE with pathologic confirmation. One patient was found to have a new PFE after 3

months suggestive of a recurrence, which to our knowledge has not been previously reported.

Patients and Methods

A retrospective review of all patients diagnosed with cardiac papillary fibroelastoma between 1996 and 2012 was performed with Institutional Review Board approval. Relevant information, including patient demographics, symptoms, characteristics of lesions, pathology, management, and follow-up were obtained.

Results

A total of 23 patients were identified who underwent surgical excision of 1 or more cardiac papillary fibroelastoma between 1996 and 2012 (Table 1). There were 13 females (57%), predominantly Caucasian (n = 16, 70%), and over 60 years of age (n = 15, 65%) with the mean age of 61.6 years. Presumptive diagnosis of PFE was made either by transthoracic (TTE) or transesophageal echocardiogram (TEE) and confirmed with histopathology. The most common cardiac comorbidities were coronary artery disease, hypertension, hyperlipidemia, diabetes, and congestive heart failure.

In 23 patients, there were 29 lesions (27 valvular and 2 muscular) (Table 2). Nineteen of the patients had solitary lesions (83%). Most lesions ranged in size from 0.5 to 1.0 cm (62%). The most common valve involved was aortic valve, followed by mitral valve.

Table 3 summarizes the presenting symptoms based on tumor location for the 23 patients. The majority of patients were symptomatic (13 out of 23, 57%) and the most common symptom was thromboembolic event (9 out of 13, 69%). All patients underwent extensive workup and the presence of an intracardiac tumor was the only etiology located. Eighteen patients (78%) had lesions on either the mitral or aortic valve. The patient with pulmonary emboli had lesions on the pulmonic valve and right atrium.

Of the 10 patients who had other cardiac symptoms the most common was angina, leading to diagnosis of either unstable angina or myocardial infarction. In addition to excision of the PFE, all these patients underwent concomitant surgical treatment for the symptomatic disease.

Three patients were completely asymptomatic. Two patients had lesions that were found on preoperative echocardiograms for cataract surgery and prostate surgery, respectively. These lesions were on the left ventricle and aortic valve, respectively. The third patient's lesion was discovered as part of annual follow-up for mitral regurgitation and was located on the aortic valve.

There was no obvious correlation between the size of the PFE and the development of symptoms. All patients with neurologic events had lesions ranging in size from less than 0.5 cm to 1 cm in diameter, and this was similar to the range in asymptomatic patients (0.7 cm to 1.5 cm).

Grossly, all lesions were described as soft, friable, yellow to yellow-white masses sometimes associated with a thin stalk. Some were described as frond or cauliflower-like in appearance. Microscopically, they had papillary projections or architecture with a hypocellular matrix and lined with endothelial cells. All pathologic specimens were confirmed to be papillary fibroelastomas.

Mean follow-up for all patients was 37 months (ranging from 1 month to 10 years). Eighteen patients had postoperative TTEs and 1 had a TEE, with evidence of a recurrent lesion in only 1 patient. Average time from surgery to follow-up echocardiogram was 18 months. There were 2 documented deaths. One was due to respiratory failure several months postoperatively; 1 occurred 7 years after surgical excision. Two individuals were lost to follow-up.

The possible recurrence was found in a 54-year-old man who presented with a past medical history significant for Hodgkins lymphoma requiring splenectomy and mantle radiation therapy in the 1970s and history of pulmonary embolus in 2003. He developed progressive dyspnea and fatigue in July 2005. He was diagnosed with right-sided pulmonary emboli by chest computed tomography (CT) scan and was managed medically with warfarin. Workup included a cardiac magnetic resonance imaging (MRI) that revealed 2 masses, 1 in his right atrium and another on the pulmonary valve. These findings were confirmed with both TTE and TEE, with lesions measuring 1×1.5 cm, and 0.8×0.5 cm, respectively. Despite appropriate anticoagulation, the pulmonic valve lesion increased in size during surveillance and his symptoms persisted. He underwent successful surgical excision of both lesions. Microscopic histology for both the pulmonary valve and right atrial lesions confirmed the diagnosis of papillary fibroelastoma.

The patient presented 3 months later with recurrence of respiratory symptoms and new bilateral lower extremity swelling. Repeat CT of the chest revealed new bilateral pulmonary emboli and a new lesion on his aortic valve, suspicious for a new papillary fibroelastoma. Echocardiograms measured the mass at 1.0×0.7 cm. He was also found to have severe aortic stenosis and constrictive pericarditis by echocardiogram and cardiac catheterization. The operating surgeon and 2 independent cardiologists reviewed all the original radiologic images including a chest CT, MRI, 2 TTEs and 2 TEEs. All concurred that the aortic valve lesion was not present in any of these original studies.

He returned to the operating room and underwent excision of the mass, aortic valve replacement, and pericardiectomy. Final pathology of the aortic valve lesion confirmed it was a papillary fibroelastoma. He was discharged home but was readmitted several months later for respiratory distress. However, the patient refused intubation and care was withdrawn.

Comment

The natural history of PFE is largely unknown due to lack of longitudinal follow-up studies. Similarly, the etiology is unknown although several hypotheses exist based on gross, microscopic, and molecular characteristics of PFE specimens [4]. Papillary fibroelastomas

have been described as neoplasms, hamartomas, organizing thrombi, or posttraumatic tumors. The organizing thrombi hypothesis is supported by the presence of fibrin, hyaluronic acid, and laminated elastic fibers within the fronds [5, 6]. The presence of miniature tendinous chords, as well as the association of congenital PFE with other congenital cardiac anomalies, provides the basis for the hamartoma hypothesis [7, 8]. Another proposed association is the relationship between PFE generation and iatrogenic trauma related to hemodynamic changes from mechanical alterations or direct endocardial damage from radiation exposure [9, 10].

Our findings are consistent with that of other investigators [11–15]. In this series, this tumor was found primarily on cardiac valves (aortic > mitral > pulmonic > tricuspid), although it can be found anywhere within the heart. Papillary fibroelastomas have also been documented within the aorta [16, 17]. The majority are solitary, less than 1.0 cm in diameter, and found in older Caucasian patients. Multiple lesions can occur at the same or different locations. One published case report documented up to 40 PFEs in both ventricles [18].

Papillary fibroelastomas can be incidental discoveries on echocardiograms, CT scans, or MRIs during cardiac catheterization or open heart surgery, or at autopsy. When symptomatic, they can present in a variety of ways, most commonly causing cardiovascular symptoms. In the majority of our patients (57%), the PFE was symptomatic with the most common being a thromboembolic event (pulmonary emboli, transient ischemic attack, cerebrovascular accident, peripheral embolus) (69%). Other documented embolic complications include retinal artery occlusion, myocardial infarction, ischemic extremity, and mesenteric ischemia [19–24]. Others report symptoms resulting from partial or complete obstruction of valves, ventricular outflow tract, or blood vessels, leading to pulmonary edema, myocardial infarction, heart failure, syncope, or sudden death [25–29]. Papillary fibroelastoma has also been associated with conduction abnormalities [30].

The management of PFE is controversial. There is no question that surgical excision is necessary in symptomatic patients with radiographically proven intracardiac tumors, regardless of the type of tumor. For patients with asymptomatic lesions undergoing cardiac surgery for other reasons, it is recommended that the PFE be excised.

In contrast, the management of asymptomatic patients remains controversial. Yee and colleagues [30] proposed a management algorithm for asymptomatic patients based on location of the PFE. Right-sided lesions could be observed and surgically excised only if they become symptomatic [30]. Left sided lesions, depending on patient comorbidities and age, could be managed expectantly, with anticoagulation or surgical excision [30]. Mariscalco and colleagues [31] proposed a slightly different algorithm, selecting characteristics of the lesion other than location as the determinant for intervention. They advise that mobile lesions, regardless of size, should be surgically excised given the higher risk of thromboembolic complications. Nonmobile lesions less than 1 cm in diameter can be managed expectantly or with anticoagulation. It is important to note that there has not been a documented case of a patient with presumed PFE that was managed successfully with anticoagulation alone. Gowda and colleagues [11] identified 25 patients with PFE who did

not undergo surgery and were managed only with anticoagulation. Twelve had PFE-related deaths, 11 of which occurred due to obstruction or embolization of coronary arteries. Four patients developed nonfatal embolization (2 coronary, 1 cerebral, 1 pulmonary). They identified tumor mobility as the only independent predictor of PFE-related death or nonfatal embolization. Although asymptomatic PFE may be observed with close echocardiographic follow-up or with anticoagulation, we recommend that surgical excision is often more prudent given the severity of potential embolization, particularly with left-sided lesions.

In addition, to our knowledge, there has not previously been a case of recurrent or metachronous PFE documented [11–15]. In the specific patient reported in this case series, his initial symptoms and pulmonary emboli could be explained by the right atrial and pulmonic valve lesions or a consequence of lower extremity deep vein thrombosis. A complete workup for an underlying hypercoagulable disorder and lymphoma recurrence was negative. Interestingly, he developed a new lesion in a different location within a 3-month period. His symptoms at the time were a combination of constrictive pericarditis physiology and bilateral pulmonary embolism. To confirm that the aortic valve lesion was not missed, all preoperative imaging was carefully reviewed by the surgeon of record and 2 independent cardiologists. All concurred that the aortic valve lesion was not visible on the original studies.

One possible explanation for this possible recurrence could be derived from the hypothesis that these lesions may have a posttraumatic origin [9, 10]. Mechanical injury related to cardiopulmonary bypass and direct manipulation of the heart causes alterations in blood flow and endothelial changes that could provide a nidus for their pathogenesis. In addition, radiation exposure could have contributed to the development of the PFE. In an autopsy series of 12 cases, Kurup and colleagues [9] suggested there was an association between the development of PFE and previous cardiac surgery or thoracic radiation. Six of these patients had previous cardiac surgery and the other 6 had thoracic radiation for breast cancer, Hodgkins, or non-Hodgkins lymphoma.

Limitations

This study is limited by the inherent bias of a retrospective study. However, given the rarity of this cardiac tumor, there have been no longitudinal follow-up studies and thus there remains limited information about the natural history of PFE.

Conclusions

Our understanding of cardiac papillary fibroelastomas began with information from autopsy series and developed with the increasing use of echocardiograms. Although rare and considered histologically benign, they can lead to serious clinical sequelae, namely thromboembolic events. Surgical management with simple excision with valvular preservation is most commonly performed. This is believed to be curative given the lack of clinical evidence of recurrence. However, we present a complex case unique for the development of a new intracardiac tumor after a 3-month period. Whether this was a recurrent or a metachronous lesion, the underlying mechanism is still speculative although approach to management is similar. There is no question symptomatic patients with

presumed PFE should undergo surgical excision. Although debate exists over the management of asymptomatic lesions, surgical excision is recommended due to the risk of thromboembolic events. Each case is unique and several factors should be taken into consideration when contemplating surgical intervention.

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Table 1**Demographics of 23 Patients With Cardiac Papillary Fibroelastoma**

Variable	Number	Percentage
Gender		
Male	10	43%
Female	13	57%
Age, years		
25–40	2	
41–60	6	
61–80	15	
Race		
Caucasian	16	
African-American	5	
Other	1	
Unknown	1	
Cardiac comorbidities		
Hypertension	16	
Coronary artery disease	7	
Hyperlipidemia	7	
Diabetes	5	
Heart failure	4	
Atrial fibrillation	3	
Complete heart block	1	
Pulmonary hypertension	2	
Cardiomyopathy	1	
Pericarditis	1	
Mitral valve prolapse	1	
Mitral valve regurgitation	1	
Mitral valve stenosis	2	
Aortic stenosis	1	
Non-cardiac comorbidities		
Osteoarthritis	3	
COPD	2	
Obesity	1	
Chronic renal failure	1	
Pulmonary embolus	1	
GERD	2	
Asthma	2	
Hypothyroidism	2	
Thyroid goiter	1	
Ulcerative colitis	2	
Hepatitis C	1	

Variable	Number	Percentage
Seizures	2	
Hodgkin lymphoma	1	
Prostate cancer	1	
Cataracts	1	
Depression	3	
Anxiety	1	

COPD = chronic obstructive pulmonary disease; GERD = gastro-esophageal reflux disease.

Table 2

Characteristics of Cardiac Papillary Fibroelastoma

Variable	Number	Percentage
Location of each lesion		
Valve	27	93%
Tricuspid	2	4%
Mitral	9	33%
Pulmonic	3	11%
Aortic	13	52%
Muscle wall	2	7%
Right atrium	1	50%
Left ventricle	1	50%
Number of lesions/patient	23	
1	19	82.6%
2	2	8.7%
3	2 ^a	8.7%
Size	29	
0.5 cm	4	14%
0.5–1.0 cm	18	62%
1–1.5 cm	6	21%
>1.5 cm	1	3%
Operation		
Excision only	11 ^b	
Excision + repair	8	
Excision + replacement	0 ^c	
Concomitant operation	10	
Excision + other	6	
CABG	1	
CABG + MVR	1	
CABG + thyroid	1	
Maze	2	
AVR	1	
Excision + repair + other	3	
CABG	2	
MVR	1	
Excision + replacement + other	1	
AVR, pericardiectomy	1 ^d	

^aThis patient had 2 lesions identified prior to first operation (right atrium, pulmonic valve). He was later diagnosed with third lesion on the aortic valve which may be a recurrent or metachronous lesion.

^bOne patient had the PFE excised from the mitral valve and its base cryoablated.

^cExcision and valve replacement refers to replacement of involved valve due to PFE.

^d Indication for aortic valve replacement was aortic stenosis despite lesion being on AV.

AV = aortic valve; AVR = aortic valve replacement; CABG = coronary artery bypass graft; MVR = mitral valve replacement; PFE = papillary fibroelastoma.

Table 3
Presenting Symptoms Categorized by Tumor Location for 23 Patients With Cardiac Papillary Fibroelastoma

Presentation	Tumor Location										Total (%)
	MV	AV	TV	PV	RA	RV	LA	LV	Multiple	Total (no.)	
Symptoms ^a											
TIA/CVA	3	1							3 ^b	7	30.4%
Dyspnea	1	1								2	8.7%
Pulmonary Embolus									1 ^c		4.3% Syncope
LE embolic event	1	1	1							2	8.7%
Incidental		2					1			3	13%
Symptoms due to other pathology ^d											
Myocardial infarction/angina	3									3	13%
Palpitations				1						1	4.3%
Mitral stenosis	1									1	4.3%
Aortic stenosis	1									1	4.3%
Goiter	1									1	4.3%
Total (no.)	5	11	2				1	1	4	23	
Total (%)	21.7%	47.8%	8.7%				4.3%	4.3%	17.4%		

^a Symptoms directly attributable to PFE.

^b One patient had 2 MV/1 TV lesions, 1 had 2 AV lesions, 1 had 2 MV lesions.

^c One patient had 1 lesion on the PV and another in the RA with recurrence of 1 lesion on the AV.

^d Presenting symptoms due to other problems; no symptoms associated with PFE.

AV = aortic valve; CVA = cerebrovascular accident; LA = left atrium; LE = lower extremity; LV = left ventricle; MV = mitral valve; PFE = papillary fibroelastomas; PV = pulmonic valve; RA = right atrium; RV = right ventricle; TIA = transient ischemic attack; TV = tricuspid valve.