State of the Art: Evaluation and Management of Patients with Cardiac Papillary Fibroelastoma

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Abstract

Objective: To review the current literature on the state of the art of evaluation and management of cardiac papillary fibroelastoma (PFE). Methods: A literature review was performed on the topic of evaluation and management of PFE. PubWeb, Web of Science, Google Scholar, Scopus search engines were used. Key search terms included: "Cardiac papillary fibroelastoma," "papillary fibroelastoma," "Surgical resection of papillary fibroelastoma," "Minimally invasive resection of papillary fibroelastoma," "Thoracoscopy and PFE." Relevant articles were archived and synthesized in an EndNote database. Particular attention was focused on identifying cases of minimally invasive resection of PFE and the use of thoracoscopic adjuncts to assist with cavitary PFE removal. Institutional Review Board approval was waived. Results: A total of 119 articles were identified and archived. Forty seven articles were used to compose this paper. 8 articles detailed minimally invasive resection of PFE. The largest series of minimally invasive PFE resection summed 4 patients. There are currently no guidelines to direct care of patients with PFE. General consensus is that the presence of left sided PFE should prompt evaluation for surgery. Conclusions: PFE is a rare tumor, more common in children than adults. Lesions are typically left sided and are at risk for embolization. Minimally invasive surgery is feasible and safe. Long-term outcomes with complete resection are very good.

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Background:

Cardiac tumors are classified by histology and site of origination. These fall into three broad categories: primary cardiac tumors, solid tumors that have spread to the heart, and infradiaphragmatic tumors that extend into the heart.[1]

Primary cardiac tumors (PCTs) are rare, with an incidence between 0.001-0.28%.[1-4] Seventy five percent (75%) are benign, with myxoma representing the most common histology. Papillary fibroelastoma (PFE) is the third most common benign cardiac tumor; but recent evidence suggests that its incidence may be higher.[5-9]

This article reviews PFE, describing tumor histology and biology, clinical manifestations, diagnostic and imaging work-up, and surgical treatment options for both valvular- and ventricular-based tumors.

Histology, Natural History, and Biology:

PFE is composed of connective tissue rich papillary fronds, covered by hyperplastic endothelium.[1] Papillary fronds provide the tumor with the classical "sea anemone" appearance (Images 1 and 2). Tumor location is predominantly left sided and valvular: aortic valve is the most common location, followed by mitral valve.[8, 10, 11] The left ventricle is the most commonly affected non-valvular site.[12]

PFE cause is unclear. There are multiple theories regarding tumor origination: microthrombi organization and consolidation, prior cardiac surgery or irridation,[13] and viral exposure.[14] Genomic analysis suggests these tumors demonstrate KRAS oncogene mutation.[15]

Clinical Manifestations:

PFE is typically discovered incidentally or following an embolic event. Embolization rates have been estimated around 35%,[16]but the true rate is unknown. PFE on the left side of the heart can cause shortness of breath, transient ischemic attacks/stroke, acute valvular dysfunction, myocardial infarction, abdominal pain and mesenteric ischemic, acute limb ischemia, pulmonary embolism, and sudden death.[5, 8, 17] Right sided PFE typically have a more insidious presentation, often remaining asymptomatic until tumor mass induces arrhythmias, pulmonary embolization, or obstructive disease.[16]

Evaluation and Imaging Work-up:

Echocardiography, cardiac MRI, and CT angiogram are all acceptable techniques to characterize cardiac based tumors.

Echocardiographic features of PFE include: small size (< 15 mm), homogenous echo dense appearance, presence of pedicle or stalk, speckled pattern, tumor edge stippling, and independent mass mobility (Images 3,4).[10, 18, 19] Tumors can appear round, oval or irregular; tumor multifocality has been reported in approximately 8% of cases.[10] PFE can be distinguished from myxoma by tumor location and echocardiographic features.[20, 21]Sensitivity and specificity of TTE at identifying PFE greater than 0.2 cm has been estimated between 88.9 and 88.4%, respectively.[10]

Transesophageal echocardiogram is an important adjunct to TTE when a cardiac tumor is identified, to delineate tumor features and anatomic involvement of intracardiac structures.[10] (Image 2). Computed tomography features of PFE include lobulated soft tissue density with a thin stalk.[22] MR findings of PFE include hypointense mobile mass on T2 weighted imaging (Image 5).[22] Challenges of both CT and MR include difficulty visualizing small tumors with cardiac motion artifact.[23]

Surgical Therapy

There are no consensus guidelines regarding PFE treatment. Due to tumor embolization risk, many groups advocate urgent resection in asymptomatic, low risk patients with left sided lesions.[8-10, 24-27] It is generally agreed upon that symptomatic patients should be offered surgery. Surgical approaches to PFE are driven by tumor location and additional concomitant surgical procedures that may be required. For solitary tumors without valvular dysfunction, simple excision is the most common resection technique.[26] Excision and valve repair/replacement has also been described with good results.

Median sternotomy historically was the most popular surgical exposure. Increasing use of minimally invasive cardiac surgery however has led to tumor removal using mini thoracotomy/Heart Port access techniques (Table 1). Intracavitary ventricular-based tumors not readily visible through surgical incisions have been resected using cardioscopy assistance (Table 2). [16-26] Cardioscopy must be performed carefully to avoid injuring the intrinsic structure of the heart.

Tumor resection principles include: complete tumor removal with preservation of cardiac structure. Cardiopulmonary bypass support is required. Procedures have been performed with and without electromechanical arrest. Recurrence rate following surgical resection is between 0 and 4%.[10, 11, 28] Recurrent disease management is driven by tumor size, symptoms, and patient performance status.

Conclusions

PFE is a rare diagnosis. Symptomatic patients should be offered surgical resection. Conservative management of small tumors should be pursued cautiously. Surgical resection should involve adherence to standard cardiac surgical principles and performance of the operation with which the surgeon has the greatest comfort. Concomitant cardiac disease should be addressed at the time of surgery. Complete tumor excision is required to reduce embolic and recurrence risk. Recurrent disease should be dealt with promptly and thoughtfully.[29]

Table 1: Minimally Invasive PFE Resection

Author	N	Tumor Location	Technique	Outcome
Hassanabad et al.[30]	1	Aortic valve	RA, mini thor	NAE.
Woo Y.J. et al.[31]	1	Aortic valve	RA, mini thor	NAE. XC 48 min

Author	**N**	**Tumor Location**	**Technique**	**Outcome**
Hua A. et al.[32]	1	Left atrium	RA, mini thor	NAE. Post-op atrial fibrillation. No recurrence 1 year.
Kim R.W. et al.[33]	1	Right atrium	Mini thor	CPB 24 min. NAE
Harling L. et al.[34]	1	Aortic valve	Mini thor	NAE. XC 16 min., discharged POD 3
Hsu V.M. et al.[35]	4	Aortic valve x 4	Partial sternotomy	NAE.
Je H.G. et al.[36]	1	LV	Mini thor, Aesop 3000 camera	NAE. XC 24 min, CPB 57 min.
Grande A.M. et al.[37]	1	Aortic valve	Partial sternotomy	NAE.

LV – Left ventricle; Mini-thor – Mini right thoracotomy; RA – Robotic assisted; NAE – no adverse events; XC – Aortic cross clamp; CPB – Cardiopulmonary bypass

Table 2: Use of Cardioscopy to Remove PFE

Author	N	Tumor Location	Technique (Optics type)	Outcome
Je, H.G. [36]	1	LV	R Mini Thor, TM (Aesop robotic)	NAE, CPB 57 min, XC 24 min.
Espada R. [38]	1	LV	TM, Stern Stryker camera 30 degree; concomitant CABG	NAE
Kaneko Y.[39]	1	LV	TM Stern, right chest thoracoscopic assistance	NAE; CPB 79 min, XC 34 min.
Misumi T. [40]	2	LV	Pt 1 – TM Stern, Olympus endoscope Pt 2 – TA Stern, Olympus endoscope	NR
Ishida K. [41]	1	LV	TA Stern EndoEye Olympus scope	NAE.
Walkes [42]	1	LV	TA Stern, 2mm thoracoscope	NAE. No recurrence 8 months.
Toeg H.D.[43]	1	LV	TA Stern, video mediastinoscope	NAE. No recurrence 1 year.
Domenech A. et al.[44]	1	LV	Transmur Stern	NAE. No recurrence 1 year.

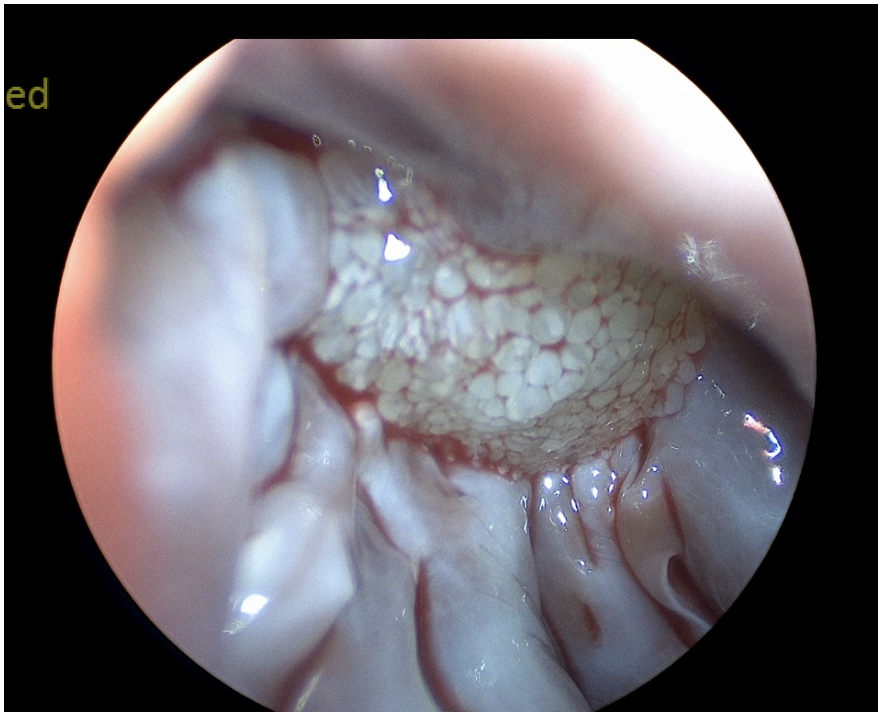
Author	N	Tumor Location	Technique (Optics type)	Outcome
Matsuo Y. et al.[45]	1	LV	TA Stern fiberoptic scope	NAE
Ariyoshi T. et al.[46]	1	LV	TM Stern, videoscope	NAE
Allen K.B. et al.[47]	1	LV	TA Stern. videothoracoscope	NAE

TM – transmitral; Stern – Sternotomy; TA – Transaortic; Mini Thor – Mini Thoracotomy; NAE – No adverse events; NR – Not reported

Image 1:

Sea anemone appearance of aortic valve PFE, when tumor placed in solution. This image demonstrates PFE post-resection. Courtesy: Neelan Doolabh, M.D.

Image 2:



Intraoperative photograph of intraventricular papillary fibroelastoma at time of surgical resection. Papillary fronds and associated left ventricular trabecular fibers can be visualized. Courtesy: John Waters, M.D. and Michael Jessen, M.D.

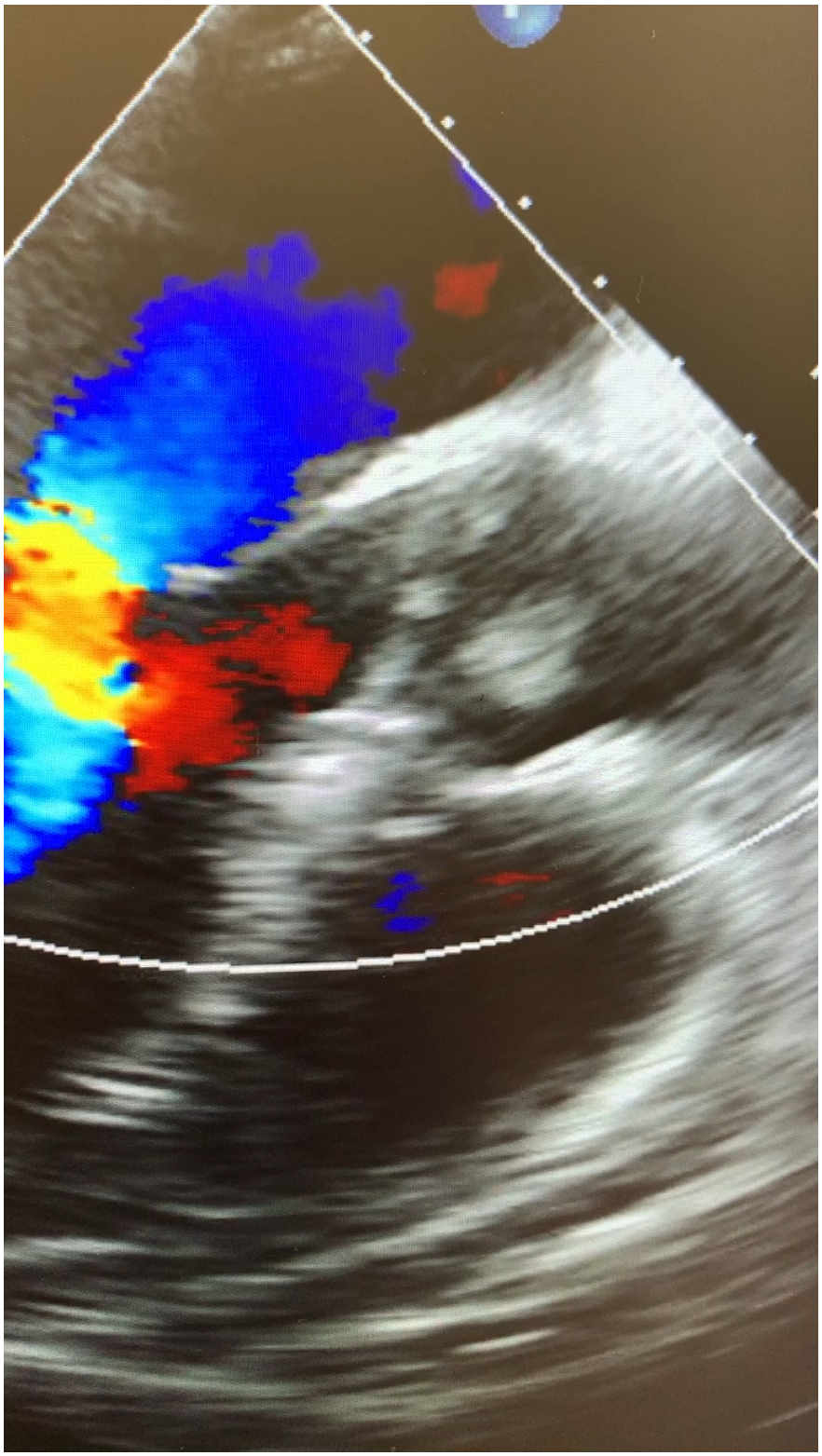


Image 3: TEE appearance of aortic valve PFE, pre-resection. Courtesy: Neelan Doolabh, M.D.

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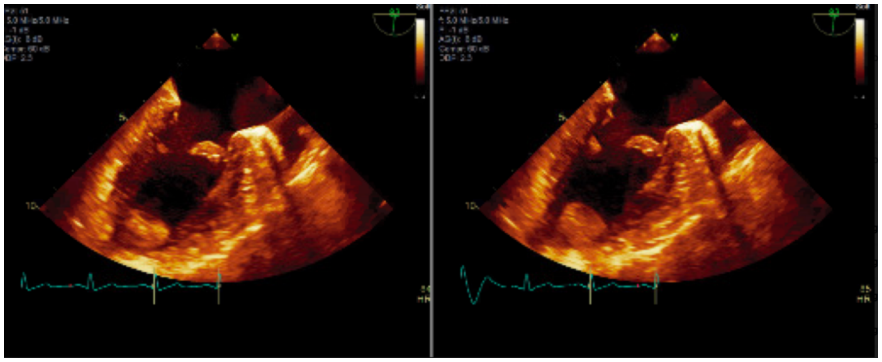

Image 4: Intraoperative TEE prior to resection, demonstrating intracavitary left ventricular mass. Courtesy: Michael Jessen, M.D.

Image 5: MR heart demonstrating 18 mm x 13 mm intracavitary PFE. Courtesy: Michael Jessen, M.D.

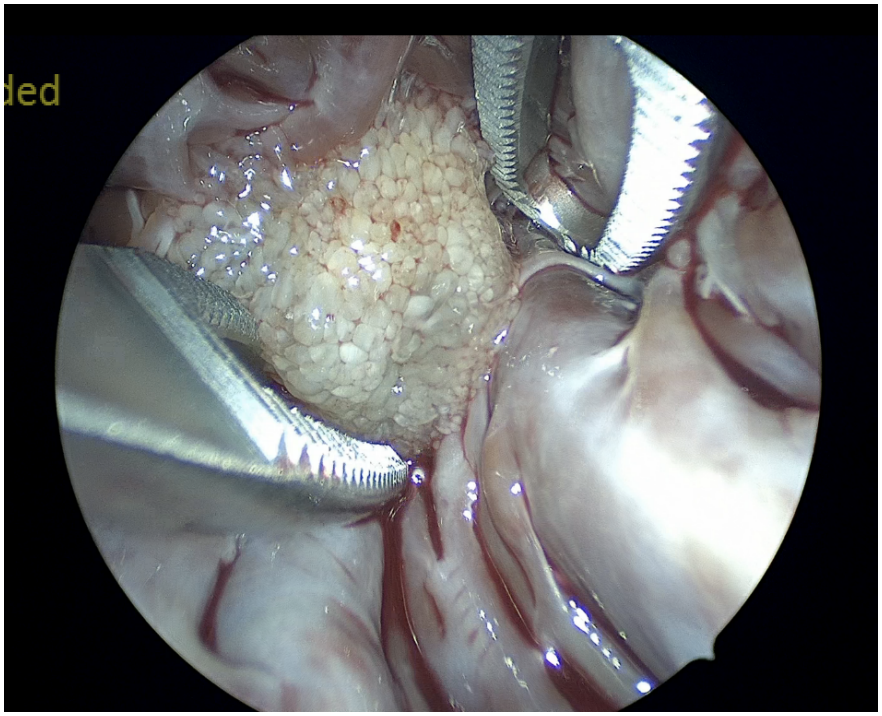

Image 6: Intraoperative photograph demonstrating cardioscopy assistance removal of PFE. 30 degree cardioscope used with mass visualization and removal. Courtesy: John Waters, M.D., Michael Jessen, M.D.

Intraoperative photograph demonstrating intracavitary fibroelastoma

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