Intrapericardial Cyst Formation in Constrictive Pericarditis Simulating Tricuspid Stenosis

By Aubrey Pitt, M.D., Robert H. Cutforth, M.D., Harvey W. Bender, M.D., J. O'Neal Humphries, M.D., George R. Stirling, M.D., J. Michael Criley, M.D., and Richard S. Ross, M.D.

SUMMARY

Two cases of constrictive pericarditis with intrapericardial cyst formation are presented. In each, a tricuspid diastolic murmur was present, and confirmation of a tricuspid valve gradient was obtained at cardiac catheterization. Angiographic studies revealed that the tricuspid valves were distorted by the cysts. In both patients, surgical excision was successfully achieved.

In case 1 a blood-containing cyst was found, the etiology of which is obscure. Case 2 was associated with rheumatoid arthritis, and the compression resulted from a chronic abscess that probably arose at a previous pericardiectomy.

Constrictive pericarditis is an uncommon disorder with a wide variety of manifestations. This communication documents two patients who presented with features of a cardiac murmur indicative of tricuspid valve stenosis. In both, exploratory surgery revealed a large cyst within scarred pericardium, narrowing the tricuspid valve. The first patient had generalized constrictive pericarditis. The second patient had undergone pericardiectomy 16 years previously for constrictive pericarditis.

Report of Cases

Case 1

A 26-year-old Negro man was admitted to The Johns Hopkins Hospital because of rapidly developing congestive heart failure. He gave no history of previous heart disease or trauma to the chest. Three weeks prior to admission he noted pleuritic pain over the anterior chest. Ten days later, when he developed a cough productive of purulent sputum, he came to the emergency room. Chest x-rays demonstrated generalized cardiomegaly. He was sent home but returned 10 days later because of ankle swelling and dyspnea of 1 day's duration.

Physical Examination

The patient was dyspneic at rest. The heart rate was 100 and regular. The blood pressure was 120/80. The arterial pulse was not paradoxical. The jugular venous pressure was elevated above the sternal angle. The apex beat was in the fifth left intercostal space outside the midclavicular line. The first heart sound was slightly accentuated. A loud third heart sound was heard over the apex of the heart, and a grade II/VI scratchy, mid-diastolic murmur which intensified significantly on inspiration was evident along the sternal border of the fourth left intercostal space. The second heart sound was narrowly split. Examination of the chest revealed no abnormality. The liver was enlarged 6 cm below the right costal margin. Pitting edema was present in the legs to the level of the knees and was also over the sacrum.

Laboratory Examination

The venous pressure was measured at 280 mm of saline. The hematocrit and white cell counts were normal. Chest x-rays revealed an enlarged globular shaped heart but no areas of calcification.

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were seen. The electrocardiogram demonstrated
generalized low voltages, flattening of the T
waves in the precordial leads, and inversion of the
T waves in leads II, III, and aVp. A tuberculin
skin test was negative. Three 24-hour
specimens of sputum did not reveal acid-fast bacilli,
and subsequent cultures were also negative. Viral
studies of the feces did not isolate Coxsackie
virus.

Table 1

<table>
<thead>
<tr>
<th></th>
<th>Preoperative (4/20/65)</th>
<th>Postoperative (7/30/65)</th>
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<tr>
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<td></td>
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<tr>
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</tr>
<tr>
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</tr>
<tr>
<td>y wave</td>
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</tr>
<tr>
<td>Mean</td>
<td>18</td>
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</tr>
<tr>
<td>Tricuspid gradient</td>
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<td></td>
</tr>
<tr>
<td>Peak early diastole</td>
<td>12</td>
<td>0</td>
</tr>
<tr>
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<tr>
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<tr>
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<td></td>
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</tr>
<tr>
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<td>18</td>
<td>4</td>
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</table>

Case 1. Scintiscan of heart with cardiac border and
diaphragm outlined. A filling defect is present within
the inferior aspect of the heart.

Scintiscans of the heart with intravenously
administered 131I-labelled iodipamide on two occa-
sions revealed decreased radioactivity in the
inferior aspect of the heart, suggesting a cardiac
tumor (fig. 1).

Cardiac Catheterization and Cineangiography
Right and left heart catheterization was
performed. The pressure data are presented in
table 1. The right ventricular end-diastolic
pressure was markedly elevated in keeping with
cardiac restriction. A pull-back tracing obtained
across the tricuspid valve demonstrated a diastolic
gradient which was most marked in the early
stages of diastole (fig. 2). The left ventricular
pressure pulse demonstrated a diastolic dip and
plateau. Simultaneously recorded left and right
ventricular pressures were nearly equal in
magnitude and configuration during diastole.

When contrast material was injected into the
right atrium, clearance was very slow. The
tricuspid valve was displaced superiorly, and the
right and left ventricles were displaced to the left.
The right atrial wall was abnormally thickened.
An injection of dye into the left ventricle revealed a small cavity and superior displacement of the floor. The right coronary artery in the atrioventricular groove was markedly displaced to the left.

These studies were interpreted as showing a mass in the heart wall displacing the cardiac chambers and causing severe distortion and narrowing of the tricuspid orifice. The etiology was uncertain, but among the diagnoses considered preoperatively were primary pericardial disease and myocardial tumor.

**Operation**

Operative exploration was performed through a median sternotomy. Exposure of the mediastinum revealed no evidence of tumor tissue. The anterior pericardium was thickened and adherent, with obliteration of the pericardial space. A mass about 6 cm in diameter was apparent on the antero-
inferior surface of the heart. An incision was made into it and 200 ml of dark-colored liquid blood escaped. This mass displaced the right atrium posteriorly and superiorly and indented the atrioventricular groove (fig. 3). The right ventricle was moved superiorly and to the left and the left ventricle assumed a more anterior position. Evacuation of the blood from the cystic space did not restore any of these abnormal displacements, and it became apparent that the entire wall of the cystic structure was composed of fibrous pericardium which did not collapse when the fluid was removed. The indention of the pericardium which remained involved the inferior vena caval-right atrial junction and the postero-diaphragmatic aspects of the heart. Frozen sections of the cyst wall showed nonspecific scar tissue with minimal inflammatory reaction. Resection of the pericardium was undertaken using sharp and pledget dissection. As the pericardium was freed from the myocardium the right ventricle bulged outward, and finally obliterated the depression which the cystic mass had produced in the heart. Complete pericardiectomy was performed removing the pericardium from phrenic nerve to phrenic nerve, dissecting it off both vessels, the right atrium, the caval-atrial junctions, the right ventricle, and a portion of the left ventricle. When the pericardiectomy was completed the heart had a normal appearance. Simultaneously recorded pressures in the right atrium and the right ventricle revealed no gradient in diastole.

Microscopic examination of the resected pericardium revealed marked fibrous thickening with chronic inflammatory changes and organizing hemorrhage. Acid-fast bacilli were not found and subsequent cultures were negative.

Postoperative Studies

The patient's postoperative course was without complication. No murmurs could be heard in the postoperative period. The patient was discharged from the hospital on the 10th postoperative day. Cardiac catheterization and cineangiography were performed 2 months postoperatively (table 1). The pressures recorded in the right side of the heart were within normal limits, and no gradient was measured on withdrawal of the catheter across the tricuspid valve. The configuration of left and right ventricular diastolic pressures was normal. Cineangiographic studies confirmed the restoration of normal relations of the heart chambers. The right coronary artery was no longer displaced.

Case 2

A 61-year-old white man was admitted to the Royal Hobart Hospital because of increasing shortness of breath of 3 months' duration. Sixteen years previously he first developed swelling and discomfort in the small joints. Five years later he was admitted to another hospital where pericardiectomy was performed for constrictive pericarditis. Postoperatively, acupuncture was required to relieve severe edema, but eventually he made a satisfactory recovery. Apart from intermittent attacks of arthritis, he remained relatively well for the next 11 years. Three months before the present admission he developed increasing dyspnea that led to his seeking further medical advice.

Physical Examination

The patient was a small, barrel-chested man who exhibited multiple arthritic deformities of a rheumatoid type affecting his hands, wrists, elbows, feet, ankles, and knees. The jugular venous pressure was elevated to the angle of the jaw. Pulsus paradoxus was present in the arterial pulse. The blood pressure was 160/90 during expiration and 134/84 during inspiration. The apex beat was in the sixth left intercostal space beneath the previous operation scar. At the lower left sternal border there was a grade I/VI pansystolic murmur and a grade II diastolic murmur which increased greatly on inspiration.

![Figure 4](http://circ.ahajournals.org/)

**Figure 4**

Case 2. Phonocardiogram illustrating diastolic murmur at left sternal border.
Table 2

*Right Heart Catheterization Data: Case 2*

<table>
<thead>
<tr>
<th></th>
<th>Preoperative (10/19/66) pressure (mm Hg)</th>
<th>Postoperative (7/28/67) pressure (mm Hg)</th>
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</thead>
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<tr>
<td>Right atrium</td>
<td></td>
<td></td>
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<tr>
<td>a wave</td>
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<td>Mean</td>
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<td>5</td>
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<tr>
<td>Tricuspid gradient</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Peak early diastole</td>
<td>10</td>
<td>0</td>
</tr>
<tr>
<td>Mean diastole</td>
<td>5</td>
<td>0</td>
</tr>
<tr>
<td>Right ventricle</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Systole</td>
<td>30</td>
<td>31</td>
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<tr>
<td>Early diastole</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>End diastole</td>
<td>18</td>
<td>4</td>
</tr>
</tbody>
</table>

(fig. 4). Auscultation of the chest revealed no abnormality. The liver was enlarged 6 cm below the right costal margin. Slight pitting edema of the ankles was noted.

*Laboratory Examination*

Pulmonary function tests revealed moderate obstruction of the airways. The hemoglobin was 12.1 g%. The white cell count was 10,400 with 90% neutrophils. The erythrocyte sedimentation rate was 21 mm in 1 hour. Liver function tests and serum electrolytes gave normal results. The latex test was negative for rheumatoid factor. Chest x-rays revealed a dense shadow in the region of the right atrium, no calcification, and normal lung fields. The electrocardiogram revealed Q waves and inversion of the T waves in leads V_2 to V_5 suggesting previous myocardial infarction.

*Cardiac Catheterization and Cineangiography*

Right heart catheterization was performed. The data are presented in table 2. Two catheters were positioned simultaneously in the right ventricle and right atrium. A diastolic gradient was present across the tricuspid valve (fig. 5). The configuration of the right ventricular pressure did not show the features of restrictive cardiac disease. During the procedure some obstruction was encountered to the free passage of the catheter at the junction of the inferior vena cava and right atrium, and some difficulty was experienced in the catheter's entering the right ventricle.

*Figure 5*

*Case 2. Simultaneous right atrial and right ventricular pressures obtained before and after surgery. A diastolic gradient is present in the preoperative recordings.*
Cineangiographic studies were carried out and at a later date angiography was repeated using a rapid film changer. A right atrial injection of contrast material was made on each occasion. These films showed the tricuspid orifice to be partially obstructed with a large filling defect in the outline of the right atrium (fig. 6).

It was uncertain whether this filling defect was due to an intracardiac or intramural mass. The patient was, therefore, transferred to the Alfred Hospital in Melbourne where cardiopulmonary bypass facilities were available.

**Operation**

The chest was opened by a midline sternum-splitting incision. The exposed pericardium was abnormal in appearance, and there was an ovoid area approximately 7 cm in diameter overlying the right atrium and part of the right ventricle, which was rather immobile and covered by a tough thickened layer of parietal pericardium. The left portion of the anterior aspect of the heart was covered by pliable normal pericardium. When the ovoid area on the right was incised, 250 cc of thick, yellow ochre inspissated material gushed out under pressure. The incision was extended from the diaphragm to the base of the heart; a very large cavity was made extending over the anterior aspect of the right ventricle and anterior and lateral aspects of the right atrium, about the inferior vena cava. The heart underlying this was covered with a shaggy granulating tissue membrane, but was quite mobile.

When the cavity was decompressed the venous pressure fell from 15 to 5 cm of water. The pericardium was removed from the anterior interventricular groove down to the phrenic nerve over the anterior and right lateral aspects of the heart. Histologic examination of the excised material did not reveal evidence of tuberculosis.

**Postoperative Studies**

Seven months postoperatively the patient was re-examined and re-catheterized. His symptoms had greatly decreased. The jugular venous pressure was normal. No cardiac murmurs could be detected, and no hemodynamic abnormality was found at cardiac catheterization (table 2). Simultaneously determined pressures in the right ventricle and right atrium demonstrated no tricuspid valve gradient (fig. 5).

**Discussion**

Auscultatory and hemodynamic evidence of tricuspid stenosis was detected in these two patients. This was associated with angiographic evidence of a mass within the heart, leading
to false diagnosis of intracardiac tumors. In the first patient there were hemodynamic changes of restrictive myocardial process, and at surgery constrictive pericarditis was found. The mass was a large blood-filled cyst distorting the tricuspid valve. The second patient had a previous pericardiectomy for constrictive pericarditis. Although there was no constriction present at the current presentation, at surgery a large cyst containing pus was found within the scarred pericardium. As in the first case, this mass distorted the tricuspid valve.

The association of a tricuspid valve gradient with constrictive pericarditis is uncommon. Eliasch and associates1 reported the finding of an early diastolic gradient across the tricuspid valve in three patients with constrictive pericarditis. No explanation was offered. Only a slight early diastolic gradient was apparent in the one case in which the right atrial and right ventricular pressures are illustrated, and this may have been due to artefactual overshoot of the right ventricular pressure. Lenzi's group2 refers to functional tricuspid stenosis possibly resulting from a tumor mass expanding into the right ventricle or resulting from obstruction to diastolic filling due to the presence of pericarditis. However, they offer no examples in their article, which is a review of the possible mechanisms of functional tricuspid stenosis. Facci and co-workers3 reported a case of chronic calcific pericarditis with tricuspid stenosis which at surgery resulted from marked restriction of the right atrioventricular groove. One case has been reported of a patient with tricuspid valve stenosis and constrictive pericarditis treated by valvotomy and pericardiectomy.4

Various valves may be involved by annular bands in patients with constrictive pericarditis. Mounsey5 reported five cases of an annular band at the level of the atrioventricular groove. In one case there was, in addition, constriction of the aortic and pulmonary valves. Other investigators have described a pericardial band producing right ventricular outflow tract obstruction6 and narrowing of the mitral orifice by pericardial retraction.7 Paul and associates8 reported three cases studied at autopsy in which a core of calcium had penetrated the left ventricle and impinged upon the mitral valve. A diastolic murmur was heard during life.

In our first case in which constrictive pericarditis was associated with a blood-filled cyst the etiology is uncertain. Trauma to the chest was suspected but emphatically denied by the patient. However, hemopericardium with resulting constrictive pericarditis has been reported to occur after mild, nonpenetrating chest trauma9,10 when the incident may have been forgotten by the patient. Tuberculous pericarditis is unlikely as there was no laboratory evidence to support this, and microscopy of the excised pericardium revealed no evidence of tuberculous lesions. Hemorrhagic effusion complicating acute non-specific pericarditis has been reported by several authors.11-14 Our patient had an episode of pleuritic pain 1 month prior to the onset of his congestive failure. Although the history of this episode is compatible with acute pericarditis, it seems unlikely that constriction would follow so quickly. Spontaneous hemopericardium has been reported to occur in male Africans by Grusin.15 Myocardial infarction may be associated with hemopericardium,16 although generalized constriction has not been reported. However, a localized hematoma may follow myocardial infarction.17

In the second case there is a clear background of constrictive pericarditis associated with long-standing rheumatoid arthritis. Lange's group18 has described a case of constrictive pericarditis associated with rheumatoid arthritis requiring pericardiectomy. Keith19 also described a case of rheumatoid constrictive pericarditis, and was able to find six other reported cases in the preceding 10 years. Gimlette20 reported five cases with rheumatoid arthritis out of a series of 62 patients with constrictive pericarditis. It seems likely that the cyst was a chronic abscess following the earlier operation of pericardiectomy.
References


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