vascular supply originating from the descending aorta above the diaphragm was secured, and the specimen was removed. A decortication was performed. The patient's hospital course was uncomplicated. He was discharged on the seventh post-operative day, and most notable was the prompt relief of pain and dyspnea postoperatively.

Pathologic Findings

The sequestered lobe measured 6 x 8 x 6 cm and weighed 60 gm. It was composed of mottled, purplish-red firm tissue with a single artery entering by the pedicle, the lumen of which was filled with a thrombus. Microscopically, it was characterized by hemorrhage and necrosis. Recognizable lung tissue was interspersed with bronchioles. The pathologic diagnosis was infarcted pulmonary sequestration.

Discussion

Extralobar pulmonary sequestration was first described in 1881 by Rokitansky and Retkowitz. Although first considered an accessory lobe, Pryce's1 clear description of intralobar pulmonary sequestration in 1946 served to consolidate the two entities. The pathogenesis remained in doubt and continues to be a source of considerable discussion. Several reviews are exhaustive in this regard.2-4 A summary of the important differences between intralobar and extralobar pulmonary sequestration is found in the Table 1 (modified from Carter5).

The most important concern of the surgeon when dealing with pulmonary sequestration is an awareness of the anomalous systemic vascular supply originating from the aorta which may arise above or below the diaphragm. Serious hemorrhage and even death have been reported when the condition is not recognized at surgery.

The treatment of extralobar pulmonary sequestration is excision. The high incidence of associated anomalies, approaching 40 percent in some series, stresses the need for complete intrathoracic exploration at the time of operation. Pulmonary agenesis, evagination or diaphragmatic hernia, ectopic pancreatic tissue, and foregut duplication or communication are the most commonly encountered entities. None was found in the present case.

References


Demonstration of Ebstein's Anomaly by Simultaneous Catheter-Tip Localization of the Tricuspid Valve and Right Coronary Artery Visualization: A New Method*

Richard E. Kerber, M.D., Melvin L. Marcus, M.D., and Paul M. Wolfson, D.O.

The diagnosis of Ebstein's anomaly has traditionally been made by angiocardiography and confirmed by simultaneous intracardiac electrocardiographic and pressure recordings. These techniques may result in false positive or negative tests. A new method is proposed, whereby the right coronary artery is used as an angiographic marker for the tricuspid annulus and a pressure catheter simultaneously marks the position of the tricuspid valve. In the right anterior oblique position the tip of the pressure catheter should be just under the right coronary artery as the tricuspid leaflets close in systole, indicating the normal relationship of the tricuspid leaflets and annulus. In Ebstein's anomaly the tip of the catheter extends well past the coronary artery in systole, demonstrating the characteristic displacement of the attachments of the tricuspid valve downward toward the right ventricular apex.

Ebstein's anomaly is a well described congenital malformation of the tricuspid valve; its characteristic feature is abnormal attachment of the tricuspid leaflets to the right ventricular wall.1-4 This can generally be demonstrated by selective right ventricular angiography.4 Intracardiac electrode catheters have also been extensively used to confirm the diagnosis.5-6 However,
several authors have reported instances of false negative results with electrode catheters.\textsuperscript{4,7,8} Watson\textsuperscript{9} also showed that false positive results could be produced in individuals without Ebstein’s anomaly, by allowing the tip of the electrode catheter to lie close to the atrial septum where there are large unbalanced electrical forces. This technique may therefore result in misleading information, delaying or preventing the correct diagnosis as in the case we report.

We suggest that by simultaneous use of an intracardiac pressure catheter and angiographic visualization of the location of the right coronary artery, the characteristic downward displacement of the tricuspid leaflets in Ebstein’s anomaly can be accurately demonstrated.

**Case Report**

A 34-year-old well developed, acyanotic man was noted to have a heart murmur at birth. He had been initially referred to the University of Iowa Hospital for evaluation in 1961, at age 21, following rejection from military service. Cardiac fluoroscopy revealed primarily right-sided cardiac enlargement, and the diagnosis of Ebstein’s anomaly was suggested. An intracardiac electrode study was performed; pullback tracings across the tricuspid valve were thought to show a normal change of the ECG complexes as the pressures changed. The diagnosis of Ebstein’s anomaly was then discarded because of this negative study (these recordings were no longer available for reinspection). The right heart pressures were normal; a small left-to-right shunt at the atrial level was suggested by oximetry, and he was discharged with the diagnosis of atrial septal defect.

In the intervening years the patient developed fatigue and exertional dyspnea, culminating in inability to work and prompting admission for reevaluation. Physical examination revealed normal vital signs and clear lungs. A mild left parasternal lift was present. S1 was increased and followed by an early systolic click appreciated best along the left sternal border. S2 was soft and not clearly split. A grade 3/6 holosystolic murmur was heard along the left sternal border; no diastolic murmurs were present. Chest x-ray examination revealed generalized cardiomegaly and normal pulmonary vasculature. The ECG showed sinus rhythm, first degree A-V block, P pulmonale, and right bundle branch block with a broad, notched r’ followed by a small s’ wave in V1. The electrical axis was superior and rightward in the frontal plane. An echocardiogram showed a marked delay in tricuspid valve closure, which occurred 0.15 second after mitral valve closure. The total amplitude of the tricuspid valve movement was 30 mm, and the mitral valve 20 mm.

These echocardiographic findings strongly suggested Ebstein’s anomaly.\textsuperscript{10–12} Because of this, cardiac catheterization was repeated. All intracardiac pressures were normal. A small bidirectional shunt at the atrial level was suggested by oximetry and Cardiogreen dye curves. Right atrial angiogram showed a very large right atrium; a large, mobile tricuspid leaflet was seen attached to the tricuspid annulus in the normal location. Additional tricuspid leaflets were thought to be visualized toward the right ventricular apex but this was not entirely clear. An intracardiac electrode catheter was then passed cephalad from the femoral vein so that it lay across the superior margin of the tricuspid valve. Pullback tracings showed that the ECG complexes changed from the ventricular to the atrial pattern simultaneously with the ventricular-to-atrial pressure change—i.e., a normal result. However, when the catheter was passed from the antecubital fossa and caudal into the right atrium and ventricle so that it lay across the inferior margin of the tricuspid valve, pullback tracings demonstrated the characteristic abnormality of Ebstein’s malformation—retention of ventricular type QRS complex after the pressure had changed to that of the right atrium.\textsuperscript{8,9} This equivocal result prompted us to delineate the relationship of the tricuspid leaflets and annulus as follows.

**Method**

A catheter was passed across the inferior pole of the tricuspid valve, pulled back just until right atrial pressure appeared, and secured in this position. The patient was then rotated to a right anterior oblique position, 30° from the horizontal; care was taken not to dislodge the right atrial pressure catheter during this maneuver. Selective right coronary arteriograms were then obtained via a percutaneous femoral approach. In this manner the position of the posterior leaflet of the tricuspid valve was marked by the pressure catheter tip, while the location of the tricuspid annulus, seen en face in this right anterior oblique view, was delineated by the right coronary artery, overlaying the annulus in the right atrioventricular sulcus.\textsuperscript{13} The catheter tip, marking the abnormally attached tricuspid leaflets, extended well past the right coronary artery toward the right ventricular apex, confirming the diagnosis of Ebstein’s anomaly (Fig 1). The procedure was then terminated.

**Discussion**

The right coronary artery runs in the right atrioventricular sulcus, overlaying the tricuspid annulus, until it divides past the acute margin of the right ventricle. It thus forms a useful angiographic marker for the location of the tricuspid annulus; by turning the patient to the right anterior oblique position the annulus and overlaying right coronary artery are superimposed. A pressure catheter positioned at the exact point of change of right ventricular to right atrial pressure will then mark the location of the tricuspid valve. In the normal individual the tricuspid leaflets, closed in systole, will lie in the plane

![Figure 1. Ebstein's anomaly, right anterior oblique position.](http://journal.publications.chestnet.org/pdfaccess.ashx?url=/data/journals/chest/20968/ on 06/26/2017)
of the tricuspid annulus and this will be denoted by the apposition of the tip of the pressure catheter and the right coronary artery (Fig 2). Although the right atrium may enlarge markedly in tricuspid regurgitation due to conditions other than Ebstein's, the anatomic relationship of the tricuspid leaflets and annulus remains undisturbed, and thus the simultaneous right coronary artery-catheter display will still appear normal (Fig 3). In Ebstein's anomaly the tip of the catheter extends well past the right coronary artery marking the tricuspid annulus, thus demonstrating the downward displacement and abnormal attachment of the tricuspid leaflet's characteristic of this condition.

There are several potential pitfalls in this technique. Care must be taken not to inadvertently dislodge the catheter positioned at the tricuspid valve when turning to the RAO position for angiography; alternatively the patient might be rotated first and then catheter withdrawal and positioning might be accomplished in the oblique position. The rotation must be sufficient so that the plane of the atrioventricular ring is at 90° to the fluoroscopic long axis of the heart and is en face to the viewer; in a PA or slight RAO position there will be overlap of the catheter and right coronary artery in normal individuals. The relative positions of the coronary artery and the catheter are best evaluated in systole; the right coronary artery displays a swinging "windshield wiper" motion in the right anterior oblique rotation and moves well to the patient's right in diastole, so that it may transiently overlap a catheter at the valve even in the normal individual. Moreover, an inadequately secured catheter might momentarily advance across the opened tricuspid valve in diastole, mimicking the Ebstein's relationship of catheter and artery.

In some cases of Ebstein's anomaly the right coronary artery is not the dominant coronary artery and may be small (Fig 1). In such cases a major branch, ie the acute marginal artery, might be mistaken for the right coronary main trunk. Such branches do not run in the atrioventricular sulcus, and attempts to relate them to the marker catheter would be misleading.

Finally, it should be remembered that in the majority of cases of Ebstein's anomaly the anterior tricuspid leaflet arises normally from the annulus; the posterior leaflet is usually displaced downward and arises below the ring while the septal leaflet is variable. Thus, the pressure catheter should be placed at the inferior pole of the valve, where the downward displacement of the leaflets is most evident, rather than at the superior pole, where the anterior and septal leaflets attach and where there may be no, or minimal, displacement. This is also supported by the variable position-dependent result of our electrode-pressure study. Correct positioning will be facilitated by passing the catheter caudad from the antecubital vein rather than utilizing the femoral vein approach; this also applies to the electrode-catheter method.

Adequate coronary visualization for the purpose of this method can often be achieved by nonselective techniques, especially in children or thin adults. Ellis et al used left atrial or left ventricular injections in two cases; aortic root injections would probably result in better coronary filling. In thin patients the radiolucent fat in the atrioventricular sulcus may be seen well enough to mark the underlying tricuspid annulus. In muscular or obese adults such as our patient, however, reliable visualization of the position of the tricuspid annulus may require selective right coronary arteriography.

We believe this method provides a specific demonstration of the characteristic anatomic abnormality of Ebstein's anomaly. Ebstein's malformation can generally be established by the use of right ventricular angiography and intracardiac electrocardiography. However, in the occasional patient in whom the diagnosis remains unconclusive after careful investigation by the traditional techniques, this new method may prove a useful and decisive diagnostic maneuver.

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Spontaneous Closure of Paravalvular Leak after Mitral Valve Replacement*

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Complications after prosthetic valve replacement may be multiple. In biologic valves, valve detachment and cusp perforation may occur. If this is of significant magnitude, reoperation may be required. This report describes recurrent mitral regurgitation after mitral valve replacement with a Hancock porcine xenograft. The regurgitation subsided spontaneously three months later. We felt that a paravalvular leak closed, with progressive fibrosis and tightening of the annulus. Functional results in this patient were excellent.

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Following mitral valve replacement, regurgitation after operation can be due to prosthetic detachment, ball variance, valve thrombosis, and in the case of biologic valves, cusp perforation or detachment. In this case, the mitral regurgitation which developed after successful mitral valve replacement with a Hancock porcine xenograft (Hancock Labs, Inc., Anaheim CA), however, subsided spontaneously three months later. We are not aware of any similar case in the literature, and therefore, present this case in some detail.

Case Report

The patient was a 38-year-old black woman who had undergone a closed mitral commissurotomy in 1961.

Physical examination revealed a chronically ill patient with findings of recurrent mitral stenosis.

The chest x-ray film showed cardiac enlargement, pulmonary vascular congestion, a small amount of pleural fluid, and left atrial and right ventricular enlargement compatible with mitral stenosis and congestive heart failure. The electrocardiogram findings were compatible with left atrial enlargement and probable right ventricular hypertrophy. There was sinus rhythm. The echocardiogram was compatible with mitral stenosis.

On April 10, 1973 the patient underwent cardiac catheterization. Cardiac angiograms revealed tricuspid insufficiency, normal coronary arteries, and no mitral insufficiency. Pressures revealed severe mitral stenosis, with a 20 mm mitral valve gradient and marked pulmonary hypertension (Table 1). On April 13, the patient underwent mitral valve replacement with a no. 29 Hancock xenograft. On exploration, the mitral valve was found to be severely stenotic and calcified.

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<th>Table 1—Catheterisation Data Before and After Mitral Valve Replacement</th>
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