

Pectus Excavatum: Abnormal Exercise Scintigraphy with Normal Coronary Arteries

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Pectus excavatum is the most common congenital abnormality of the chest wall, and is frequently associated with chest pain. The invasive, as well as the ECG and echocardiographic assessment of possible coronary artery disease (CAD) in adults with moderate to severe forms of this deformity, is often complicated by the associated displacement of the heart in the chest cavity in these patients. We present findings in a 67-yr-old male that demonstrate that the predictive accuracy of positive stress radionuclide ventriculogram (RVG) and SPECT scintigraphic studies may be significantly reduced in patients with moderate to severe forms of this abnormality. Our findings also suggest, however, that either lateral or even a shallow left posterior oblique detector positioning during RVG, a significantly revised SPECT acquisition orbit, or planar imaging may provide a more accurate means to assess possible CAD in these patients. Likewise, physician input would appear to be invaluable in determining the optimal mode of imaging and the acquisition protocol for patients with pectus excavatum.

Key Words: pectus excavatum; coronary artery disease; radionuclide ventriculogram

J Nucl Med 1994; 35:1985-1988

Pectus excavatum is the most common congenital abnormality of the chest wall (1). Thirty to seventy percent of patients who have this deformity present with symptoms of chest discomfort, dyspnea on exertion and exercise intolerance, is often related to the chest wall abnormality itself (2). In addition, pectus excavatum occurs along with other conditions often associated with chest pain, such as idiopathic mitral valve prolapse and Marfan's syndrome (1,2). Coronary artery disease can also occur in adult patients with pectus excavatum. However, the noninvasive evaluation of possible CAD can be complicated by the baseline electrocardiographic (ECG) abnormalities frequently observed in patients with this abnormality (2,3). Specifically,

patients with moderate to severe forms of this chest wall deformity, anterior Q-waves on ECG often represent a pseudoinfarct pattern that results from posterior and lateral displacement of the heart in the chest cavity (2), thereby complicating the interpretation of both the resting and exercise ECG. Likewise, exercise echocardiography is often technically difficult in these patients due to the chest wall deformity and associated displacement of the heart in the chest. Exercise scintigraphy can be used to attempt to increase the reduced specificity and sensitivity of exercise testing in these patients (2,3), but the diagnostic accuracy of exercise scintigraphy in this setting is unknown. In this report, we present the first description of pectus excavatum as a cause for positive exercise scintigraphy in a patient with chest pain and angiographically normal coronary arteries and normal coronary flow reserve. The findings presented suggest that the predictive accuracy of positive exercise scintigraphic studies may be significantly reduced in patients with this chest wall deformity unless careful attention is given to the mode of acquisition and the imaging protocol used.

CASE REPORT

A 67-yr-old male with pectus excavatum was evaluated in February 1993 when he presented with several episodes of chest pain, one of which prompted a visit to the emergency room. Physical examination revealed a thin, healthy appearing man in no distress, with a blood pressure of 125/75 and a regular heart rate of 64. Examination of the thorax revealed severe pectus excavatum. The lung fields were clear to auscultation. On cardiac examination, the apical impulse had a normal quality, but was displaced leftward toward the axilla. The first and second heart sounds were normal. There was a grade I/IV short, early systolic murmur heard at the upper left sternal border without radiation. The remainder of the physical exam and routine laboratory tests were normal. His ECG revealed a sinus rhythm with a first degree AV block and Q waves in leads V₁-V₄. A technically difficult echocardiogram and Doppler flow study demonstrated only mild myxomatous changes of the mitral valve without prolapse, trace aortic insufficiency and normal left ventricular (LV) function. Due to the patient's chest wall deformity, nonstandard views were utilized during the echo acquisition, precluding an accurate determination of LV and right ventricular (RV) dimensions.

Received Feb. 8, 1994; revision accepted Aug. 8, 1994.

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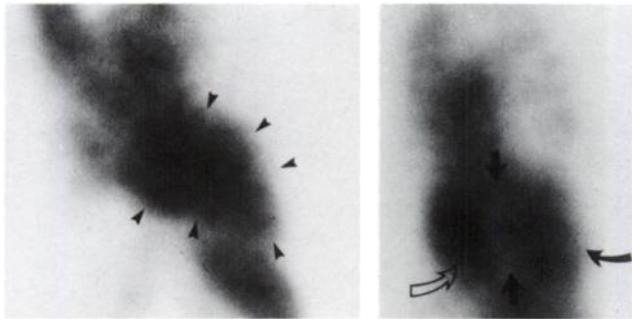


FIGURE 1. Resting radionuclide ventriculograms (RVGs) obtained using the standard LAO 45° gamma camera position (left) and a nonstandard left lateral gamma camera position (right) in the same patient. The RVG on the left appears to demonstrate a large and elongated LV (outlined by closed symbols), actually created by overlap of both the RV and LV blood pool images. The RVG on the right clearly shows the interventricular septum (straight arrows), with distinct RV (open, curved arrow) and LV (closed, curved arrow) images.

To evaluate his symptoms, an exercise radionuclide ventriculogram (RVG) was performed using a supine bicycle. The test was stopped secondary to fatigue at 9 min of exercise, after reaching a peak heart rate of 150 and a rate-pressure product of 24,000. The patient developed 1 mm of upsloping ST depression in the inferior leads. Imaging was performed using a Siemens Orbiter single-headed gamma camera (Siemens Medical Systems, Inc., Hoffman Estates, IL) with a low-energy, general purpose parallel-hole collimator and an energy window of 20% centered at 140 keV. An in vitro method was used to label the patient's red blood cells with 30 mCi of ^{99m}Tc . The patient was studied in the semi-erect position with the imaging field of view centered on the heart. Best septal separation of the right and left ventricles was attempted by moving the gamma camera from the anterior to the nearly left lateral position, with the best blood pool separation thought to be obtained in the LAO 45 position. A 2-min baseline study was acquired prior to the onset of exercise. Exercise acquisitions were obtained during the final 2-min period of each 3-min exercise stage. The patient did not exhibit an unusual degree of movement during the exercise stages and a motion algorithm was not employed. Background regions of interest (ROIs) were determined to be satisfactory and the ejection fraction of the apparent left ventricle was calculated to be 61%. Reconstructed images, however, showed the left ventricle to have an elongated and prominent appearance, with nearly complete obscuration of the right ventricular blood pool (Fig. 1). In addition, the patient was found to have an abnormal LV ejection fraction (LVEF) response to exercise, with the LVEF changing from 74% during stage I, to 71% during stage II and finally to 63% at peak exercise. He subsequently underwent further noninvasive evaluation with a stress thallium study. He exercised to a peak heart rate of 144 and a peak rate-pressure product of 21,024 with similar ECG changes. Three millicuries of ^{201}Tl -chloride were intravenously administered 1 min prior to the termination of exercise. SPECT thallium images were obtained with a Siemens Orbiter single-headed camera equipped with a 3/8-inch crystal and a low-energy, general purpose parallel collimator. A 25% window centered at 75 keV and a 20% window centered at 167 keV were utilized. Imaging onset was within 10 min after termination of the ECG stress study. An 180° elliptical orbit was utilized beginning at 45° RAO and ending at 45° LPO. Thirty-two images were acquired using a "step and

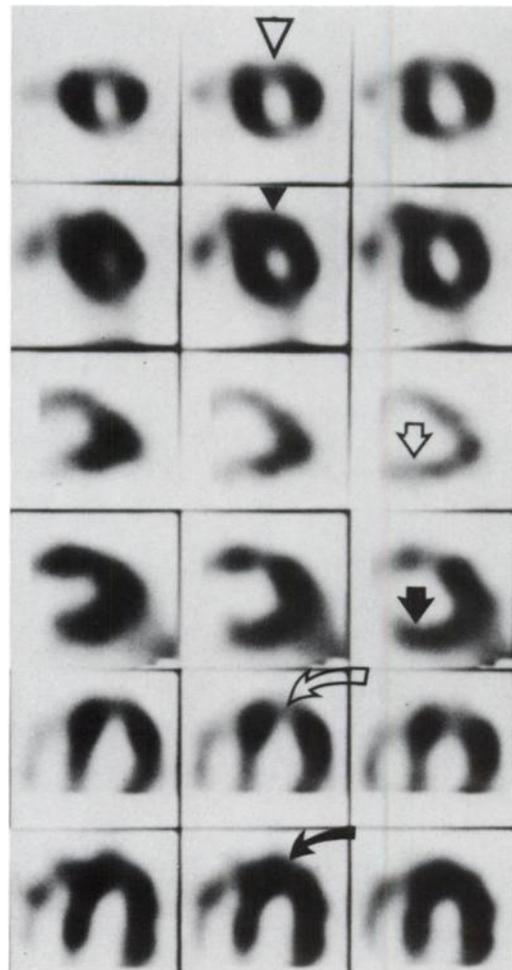


FIGURE 2. SPECT thallium images, with stress images above rest images, demonstrating short-axis (top 2 rows), vertical long-axis (middle 2 rows) and horizontal long-axis (bottom 2 rows) views. Multiple reversible perfusion abnormalities are evident. Examples of stress defects (open symbols) and their resolution on rest images (closed symbols) are pointed out in the anterior wall (triangles), inferior wall (straight arrows), and apex (curved arrows).

shoot" acquisition mode at 6-degree intervals. A 64×64 matrix was used with a Butterfield filter frequency cutoff of 0.45. After the initial images were obtained, 2.0 mCi of ^{201}Tl were injected and delay SPECT images were obtained 4 hr later. Images were checked for patient motion and a motion correction algorithm was not employed. Following selection of the long-axis of the left ventricle, tomograms of 1 pixel thickness were obtained in transverse, sagittal and coronal orientations. SPECT images were remarkable for multiple moderately to severely reversible perfusion abnormalities (Fig. 2), consistent with severe multivessel CAD.

Because of the patient's symptoms and these abnormal exercise scintigraphic studies, a diagnostic cardiac catheterization was performed. Cardiac fluoroscopy at the time of the catheterization demonstrated marked rotation of the heart, with the LV apex markedly displaced, both posteriorly and to the left. Left ventriculography revealed normal end systolic and end diastolic dimensions, with a normal LVEF. Although rotation of the heart and aortic root made coronary angiography technically difficult, the epicardial coronary arteries were selectively cannulated and were normal. In addition, coronary artery flow reserve was evaluated

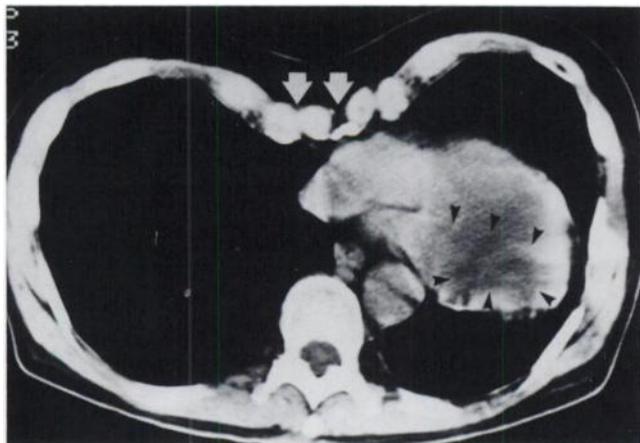


FIGURE 3. Chest CT scan demonstrates severe pectus deformity (white arrows) with marked lateral and posterior displacement of the LV (outlined by dark symbols) in the chest cavity.

using a 3F Doppler catheter. At a paced heart rate of 100, the coronary flow reserve was found to be within normal limits.

Once the cardiac anatomic information was available, we repeated the RVG with a shallow LPO acquisition to determine if correction for displacement of the heart within the chest wall would provide a more accurate assessment of changes in left ventricular function with exercise. This study demonstrated normal separation of the LV and RV, with clear visualization of the interventricular septum (Fig. 1). The patient again performed supine bicycle exercise. The resting LVEF was 66%. During exercise, this changed from 65% to 74%, and finally to 79% at peak exercise; a normal ejection fraction response.

DISCUSSION

Pectus excavatum has an estimated incidence of 0.8% in the general population (1). Although this disorder appears to occur sporadically, a familial tendency for its occurrence has been noted (2). The severity of the sternal deformity in pectus excavatum ranges from an almost unnoticeable narrow central cleft to a broad dish-shaped defect that reaches the spine in the most severe form. Many methods to assess the severity of this abnormality have been proposed but none have gained universal acceptance. Fabricius et al. used the sternovertebral distance on the chest x-ray to classify the defect as mild (>7 cm), moderate (5–7 cm) or severe (<5 cm) (4). Others have advocated using the ratio of the transverse diameter-to-the most narrow anterior-posterior (AP) diameter on CT scans of the chest to determine the severity (5). If the ratio is greater than 3.25, the defect is considered severe and surgical repair may be recommended. A CT scan of the chest in the patient described above (Fig. 3) demonstrated that the smallest AP diameter was 5.8 cm and the transverse to AP ratio was 4.7, indicative of a moderately severe deformity. The CT of the chest in this patient also demonstrated marked posterior and lateral displacement of the heart (Fig. 3), which occurs in moderate to severe forms of this chest wall deformity.

Patients with pectus excavation have previously been

noted to have abnormal ECG responses to exercise, despite normal epicardial coronary arteries on angiography (3). To our knowledge, however, this is the first reported case that demonstrates an association between pectus excavatum and false-positive exercise scintigraphy. The initial exercise RVG in the patient described above was clearly abnormal, yet coronary flow reserve and arteriography were both normal. Although a significant reduction in the LVEF observed on RVG during exercise is consistent with myocardial ischemia (6), the specificity of this response may be reduced in a number of situations (7–9). Our patient's false-positive RVG appears to be related to his severe pectus abnormality and the camera angle used during image acquisition. This was suggested by the enlarged and elongated LV image obtained on the RVG (Fig. 1), incongruent with the normal LV size observed on left ventriculography at the time of the patient's cardiac catheterization. Likewise, the interventricular septum and normal separation of the LV and RV images were never clearly demonstrated on the initial RVG (Fig. 1). Due to the profound lateral displacement of the apex of the patient's heart (Fig. 3), there was apparent separation of blood pools on the baseline RVG, which inadvertently resulted in improper positioning of the gamma camera by the technologist and physician involved in this patient's image acquisition. In retrospect, the LAO 45° position, which was initially thought to be the best septal image, provided good atrioventricular separation but poor ventricular separation. Ventricular separation would have been better obtained in an extremely unusual shallow LPO orientation (Fig. 1). Thus, it would appear that patients with moderate to severe pectus excavatum may be particularly susceptible to false-positive exercise RVG studies unless the gamma camera position is extremely adjusted to a lateral or extreme LPO position to compensate for the marked displacement of the heart in these patients.

We were able to find only one previously published report that used RVG to evaluate the LVEF response to exercise in patients with pectus excavatum (10). However, that study was performed in children who had only mild to moderate pectus excavatum and not in adults with moderate to severe forms of this abnormality. Moreover, although there was an overall increase in the mean LVEF from rest to exercise in the group of subjects evaluated in that study, individual responses were not reported (10). Thus, it is possible that some of the individuals with mild to moderate pectus deformity in the evaluation by Peterson et al. may have also demonstrated abnormal LVEF responses to exercise despite the normal mean response of the entire group.

The patient described in our report also had a false-positive SPECT thallium exercise study. A review of the literature revealed only one other study that has reported the results of thallium imaging in patients with pectus excavatum (11). The objective of that study was to evaluate the etiology of chest pain in patients with mitral valve prolapse syndrome. In that report by Gaffney et al., three

of the patients with mitral valve prolapse also had pectus excavatum and were found to have normal exercise thallium images and angiographically normal epicardial coronary arteries (11). Unfortunately, the severity of the pectus deformity in these patients was not reported. In addition, planar imaging was used in that earlier study, instead of SPECT imaging more commonly used at this time. Nonetheless, these very limited findings would suggest that the predictive accuracy of a negative thallium study may not be reduced in patients with pectus excavatum. By contrast, the SPECT thallium study of the patient described in our report was clearly abnormal, demonstrating multiple reversible perfusion abnormalities (Fig. 2). Although the results of this patient's exercise study were strongly suggestive of multivessel CAD, his coronary angiogram was normal. Similar abnormal findings on exercise thallium studies can also be seen in patients with hypertension, LV hypertrophy (LVH), and in patients with microvascular disease secondary to other etiologies (12-14). However, the patient described in our report did not have hypertension or LVH. He also had normal coronary flow reserve, excluding the possibility of significant small-vessel disease. Therefore, the SPECT thallium study in this patient was, in fact, a false-positive study.

Others with moderate to severe forms of pectus excavatum would also appear to be especially susceptible to false-positive thallium studies using standard orbit acquisitions, due to the abnormal position of the heart in these patients. Lateral displacement of the heart in the chest cavity results in significant artifact on SPECT thallium images (14,15). The standard 180° gamma camera arc used in ²⁰¹Tl SPECT imaging does not place the heart in the center of the camera orbit when the heart is significantly displaced laterally. Resulting artifact is created by differences in spatial resolution that occur when the heart-to-detector distance changes during imaging (15). Furthermore, SPECT images are also subject to substantial artifact when thallium count densities are significantly attenuated (12,14), as can occur when posterior displacement of the heart in the chest results in a substantial increase in the distance between the detector and the heart or when the sternum directly overlies the image of interest. As a consequence of both lateral and posterior displacement of the heart in patients with moderate to severe forms of pectus excavatum, resultant artifacts may occur on stress and rest SPECT thallium images when a standard imaging orbit is used. Thus, these patients may demonstrate reversible and/or fixed SPECT thallium perfusion abnormalities that are very suggestive of significant CAD despite having normal coronary arteries, as was the case for the patient presented in this report.

Therefore, the predictive accuracy of positive SPECT

thallium studies, as well as positive exercise RVGs may be significantly reduced in patients who have pectus excavatum and associated displacement of the heart, unless careful attention is given to the imaging protocol used. Likewise, stress echocardiography may be suboptimal in these patients due to poor image quality associated with displacement of the heart. As a consequence, exercise planar thallium imaging, a revised SPECT acquisition orbit, or RVG with imaging in either the left lateral or shallow LPO position would appear to be preferred modalities to noninvasively assess possible ischemic heart disease in patients with moderate to severe pectus excavatum. Thus, physician input in determining the optimal acquisition protocol during exercise scintigraphy would appear to be invaluable in overcoming the problems resulting from cardiac displacement in patients with pectus excavatum.

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