

Pectus excavatum: echocardiography and cardiac MRI reveal frequent pericardial effusion and right-sided heart anomalies

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Aims

In patients with pectus excavatum (PEX), echocardiographic assessment can be difficult. There are little data on the impact of the chest deformity on echocardiographic findings and comparison of data obtained by echocardiography (echo) with cardiac magnetic resonance imaging (CMR) in PEX.

Methods and results

In a prospective study, cardiac anomalies in PEX were analysed by echo and compared with CMR in consecutive patients with PEX referred for echo. If they agreed to participate, the patients were referred for CMR and included if the pectus index was ≥ 3.0 by CMR. Also, clinical data and electrocardiogram tracings were analysed. There were 18 patients (13 females; 72%), with a mean age of 53 ± 16 years; mean pectus index was 4.7 (range: 3–7.3). Echo showed haemodynamically insignificant pericardial effusion in six patients (33%), tricuspid valve prolapse in five (28%), right ventricular (RV) localized wall motion anomalies (WMA) in five (28%) and diminished RV systolic function in two (11%); no patient had RV dilatation. CMR demonstrated cardiac displacement to the left in 9 patients (50%); minimal pericardial effusion was seen in 10 patients (56%; P value = 0.13 compared with echo), RV localized WMA in 6 (44%; P value = 1.0), diminished RV systolic function in 8 (44%; P = 0.07), and RV dilatation in 5 (28%; P = 0.06). A completely normal cardiac examination was found in six patients by echo (33%) and in 2 (11%) using CMR. Although some signs of arrhythmogenic RV cardiomyopathy (ARVC) were present, no patient fulfilled the ARVC criteria.

Conclusion

In severe PEX, haemodynamically insignificant pericardial effusion, tricuspid valve prolapse and other RV anomalies possibly due to RV displacement are frequent as demonstrated by both CMR and echo. The cardiac assessment by echo and CMR did show discrepancies; however, they were not significant.

Keywords

Pectus excavatum • Pericardial effusion • Right ventricle • Arrhythmogenic right ventricular cardiomyopathy • Echocardiography • Cardiac magnetic resonance imaging

Background

Pectus excavatum (PEX) is a common deformity of the chest wall in which the inferior part of the sternum and the cartilage are displaced posteriorly. PEX occurs in $\sim 1:400$ to $1:1000$ live births with a male-to-female ratio of 4:1.^{1–3} PEX has been described in

scoliosis, congenital heart disease and syndromes such as Marfan syndrome or Noonan syndrome⁴ or in other conditions such as mucopolysaccharidosis. Children and adolescents rarely have symptoms due to PEX but may suffer from the cosmetic impact of this chest deformity; older patients may complain about lack of endurance, syncope, dizziness, dyspnoea on exertion, anterior

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chest pain and palpitations.⁵ The chest deformity in PEX may lead to compression of the right-sided cavities between the sternum and the vertebral column⁵ impacting right ventricular (RV) anatomy and rendering cardiac assessment by echocardiography very difficult. Besides, in every day practice we noticed a higher incidence of echofree spaces in patients with PEX which had never been analysed in detail by cardiovascular magnetic resonance (CMR) imaging.

The aim of our study was to describe cardiac findings in PEX found by echocardiography and to compare these with CMR.

Methods

Study subjects

Patients with PEX undergoing clinically indicated cardiac evaluation by transthoracic echocardiography and standard 12-lead electrocardiogram (ECG) were prospectively recruited for this study. Inclusion criteria were age >18 years and a pectus index of ≥ 3 at cross-sectional imaging by CMR. Exclusion criteria were previous funnel chest operation or previous cardiac surgery. Between January and December 2009, 21 patients consented to participate in the study and to undergo CMR as an additional examination. Three patients were excluded as their pectus index was <3 by CMR leaving 18 patients in the study. Clinical data including functional class [New York Heart Association (NYHA)], history of palpitations, arrhythmias, oedema, and/or syncope were also obtained.

The study was approved by the ethical committee of our institution; a patient consent form was signed by each patient.

Echocardiography

Complete transthoracic echocardiography was performed according to previously published guidelines.⁶ Age-adjusted assessment of diastolic function was performed as recommended by Nagueh *et al.*⁷ In addition to the usual parameters, the following echocardiographic measurements were performed in all patients: RV outflow tract (RVOT) and fractional area change of the right ventricle (FAC; normal >32%). The following echocardiography machines were used: Vivid EchoPac PC v.7.0.1 (GE Healthcare, Horten, Norway) or Philips IE 33 (Philips Medical Systems, Bothel, WA).

The RVOT was considered to be dilated if >29 mm measured above the aortic valve.^{6,8} FAC was measured in the apical four-chamber view; its normal value was 32–60% as previously described.⁶ Regional zones of the myocardium thinned and/or akinetic, hypokinetic, or aneurysmatic, respectively, dyskinetic were called localized RV wall anomalies.⁸

Cardiac magnetic resonance imaging

All CMR examinations were performed using a 1.5-T scanner (Signa HDx, General Electric Medical Systems, Milwaukee, WI, USA) and an 8-channel phased-array cardiac coil. The images were acquired during breath holding in all patients, by asking the patients to stop breathing in end inspiration.

The protocol started with a triplane localizer with non-gated cine steady-state free precession (SSFP) sequences. Gated SSFP images were then acquired in a vertical and horizontal long-axis plane, in a stack of short-axis acquisitions covering the entire length of both ventricles, and in an oblique plane through the RVOT.

Image analysis

The pectus index (Haller index)⁹ was measured on axial images and defined as the ratio of the maximum internal transverse diameter of the thorax to the minimum sternovertebral distance.

Cardiac displacement was assessed by tracing a sagittal line between the sternum and the spine; in normal subjects this line passes through the right ventricle anteriorly and the left atrium posteriorly. The heart was considered displaced if only one or both atria were observed in the space between the sternum and the spine.

The measurement of the ventricular volumes and function was performed on a commercially available off-line workstation (Sun Microsystems, Inc., Mountainview, CA, USA) using the Mass+ Software package (Magnetic Resonance Analytical Software System, Version 4.0, MEDIS, Medical Imaging Systems, Leiden, The Netherlands) as previously described.^{10–12} The normal values for the left- and the right ventricles recently published by Maceira *et al.*^{10,11} were taken as the reference for defining the ventricular dilatation.

Pericardial effusion was defined as the presence of a bright layer >4 mm between the epicardium and the pericardium on the SSFP images, and of a line of low signal intensity between the internal epicardial fat and the external pericardial fat on the fast spin echo images, both on a short axis and horizontal long projection.¹³ The RV myocardium was assessed on black blood images for thickness and by suspected presence of fat infiltration. Regional hypokinetic areas, bulging or microaneurysm were evaluated on the SSFP axial images and on the images showing the RVOT. The RVOT was considered dilated if its diameter was >29 mm.^{14,15}

Diagnostic criteria for ARVC

The revised diagnostic criteria for ARVC by 2D echo, MRI, and RV angiography, tissue characterization, analysis of repolarization abnormalities, depolarization/conduction, arrhythmias and family history have been defined by the Task Force of Cardiomyopathies.¹⁶ Major echocardiographic criteria include regional RV akinesia, dyskinesia, or aneurysm and one of the following: regional RV akinesia or dyskinesia and one of the following: RVOT dilatation in the parasternal long (≥ 32 mm) or short axis (≥ 36 mm), or FAC $\leq 33\%$. Major criteria by CMR include regional RV akinesia or dyskinesia or dyssynchronous RV contraction and one of the following: ratio of RV end-diastolic volume to BSA ≥ 110 mL/m² (male) or ≥ 100 mL/m² (female) or RV ejection fraction $\leq 40\%$. Major ECG criteria include inverted T waves in right precordial leads (V1–V3) or beyond in individuals >14 years of age (in the absence of complete right bundle-branch block QRS ≥ 120 ms) and epsilon waves (reproducible low-amplitude signals between the end of QRS complex to onset at the T wave) in the right precordial leads (V1–V3). For definite diagnosis two major or one major and two minor or four minor criteria from different categories have to be fulfilled.¹⁶

Statistical analysis

Values are presented as mean \pm standard deviation. Categorical variables are presented as frequencies and percentages. Comparisons between groups were performed by a Wilcoxon signed-rank test in continuous variables and the McNemar test for categorical variables.

Results

Clinical data are summarized in Table 1. Due to the significant PEX, cardiac symptoms were frequent including dyspnoea on exertion, palpitations, syncope, and/or chest pain occurring in 15 patients (83%). However, only one patient (6%) was in functional class

Table 1 Clinical characteristics in the 18 subjects with PEX

Age (years)	53 ± 16
Female gender	13 (72%)
Height (cm)	170 ± 8
Weight (kg)	61 ± 10
BSA (m ²)	1.69 ± 0.16
Functional class	
NYHA I	9 (50)
NHYA II	8 (44)
NYHA III	1 (6)
NYHA IV	0 (0)
Dyspnoea on exertion	9 (50)
Palpitations	9 (50)
Chest pain	2 (11)
Syncope	4 (22)
Oedema	1 (6)

Percentage values are indicated in parenthesis. BSA, body surface area; NYHA, New York Heart Association.

Table 2 Summary of ECG findings

Sinus rhythm	18 (100)
Heart rate (bpm)	69 ± 12
Supraventricular arrhythmias	10 (56)
Epsilon wave	0 (0)
Inverted T wave/s V1–V3	0 (0)
Right bundle-branch block	
Incomplete	10 (56)
Complete (QRS > 120ms)	1 (6)
First degree AV block	3 (17)
Delayed R/S alteration in V4–V5	8 (50)

Percentage values are given in parenthesis.

III and no patient in class IV. No patient had the Marfan syndrome, the Noonan syndrome, or other known genetic disorder.

The ECG findings are summarized in *Table 2*. Ten patients had a history of supraventricular arrhythmias. No patient had a history of ventricular tachycardia or frequent ventricular premature contractions with a left bundle branch block morphology or other morphology. The most frequent ECG abnormality was an incomplete right bundle-branch block and/or a delayed R/S transition in V4–V5 (50% each). No patient fulfilled the ARVC criteria by ECG.

The data of echocardiography and CMR are summarized in *Tables 3* and *4*.

Left ventricular size, systolic, and diastolic function were normal apart from one patient where left ventricular ejection fraction was reduced to 47%. RV end-diastolic area was normal in all as well as tricuspid annular systolic plane excursion. The RV systolic function assessed by FAC was diminished in two patients (11%). Only three patients had a mild pulmonary artery hypertension of systolic pulmonary artery pressure of >35 mmHg. Valvular

Table 3 Summary of echocardiographic findings

Parameter		Range
LVEDD (mm)	44 ± 4	38–54
LVEF (%)	62 ± 7	47–78
Abnormal diastolic function	0	
RV area, end diastolic (cm ²)	16 ± 4	10–22
RV fractional area change (%)	45 ± 10	28–64
Tricuspid annular plane systolic excursion (mm) (14 patients)	23 ± 5	18–33
Systolic pulmonary artery pressure (mmHg, 14 patients)	24 ± 8	15–41
Pulmonary artery hypertension (in 14 patients)	3 (21)	
Mitral regurgitation		
None, mild	18	
Moderate, severe	0	
Tricuspid regurgitation		
None, mild	17	
Moderate, severe	1	
Atrial septal defect	1 (6)	
Other congenital heart disease	2 (11)	

Percentage values are given in parenthesis. LVEDD, left ventricular enddiastolic diameter; LVEF, left ventricular ejection fraction.

heart disease was observed in eight patients and included tricuspid valve prolapse in five patients (only one with moderately severe tricuspid regurgitation), bicuspid aortic valve in one patient, mild mitral regurgitation in one patient, and mitral valve prolapse in one patient.

By echocardiography, haemodynamically insignificant pericardial effusion was observed in six patients (33%; see *Figures 1* and *2*). RV abnormalities as described in ARVC were present in seven patients (39%), and consisted of localized hypokinetic, akinetic, or dyskinetic areas in four patients (*Figure 3*), dilatation of the RV apex (*Figure 3*) and and/or thinning of the RV free wall in one patient each. No patient had the RV dilatation, whereas the left atrium was dilated in one patient (6%) and the right atrium in two (11%). The mean size of the RVOT was 28 ± 5 mm; a dilated RVOT was found in five patients (28%) and measured 32–39 mm. The average RV FAC was 45 ± 10% and the RV end-diastolic diameter was 2.7 ± 0.5 cm.

At CMR a mean pectus index of 4.7 ± 1.5 (range: 3.0–7.3) was measured. Significant cardiac displacement into the left hemithorax was observed in nine patients (50%; *Figure 4*).

A comparison of findings of echocardiography and cardiac MR is shown in *Table 4*. The left ventricular size tended to be slightly larger ($P = 0.08$) and the left ventricular ejection fraction slightly lower ($P = 0.05$) by CMR. The RVOT enlargement tended to be more rare by CMR ($P = 0.13$) and occurred in only one patient with CMR (34 mm). Pericardial effusion was present in 6 patients by echocardiography and in 10 patients by CMR (56%) ($P = 0.13$), and occurred not only anteriorly in front of the right ventricle, but was diffuse and distinguishable also along the posterior wall of the left ventricle in all cases (*Figure 5*). Mild RV dilatation

Table 4 Comparison of echocardiographic findings and CMR

	Echocardiography	CMR	P value
RVEDV (mL)		77 ± 14	
RVEF (%)		59 ± 7	
LVEDV (mL)	69 ± 18	77 ± 14	0.08
LVEF (%)	62 ± 7	59 ± 7	0.05
RVOT enlargement	5 (28)	1 (6)	0.13
Localized RV wall anomalies	5 (28)	6 (33)	1.0
RV enlargement	0	5 (28)	0.06
Pericardial effusion	6 (33)	10 (56)	0.13
Diminished RV systolic function	2 (11) (FAC ≤32)	8 (44) (RVEF ≤52)	0.07
Prominent moderator band	5 (28)	4 (22)	1.0
Any sign of ARVC	7 (39)	10 (56)	0.45

Percentage values are given in parenthesis. LVEDV, left ventricular end-diastolic volume; RVEF, left ventricular ejection fraction.

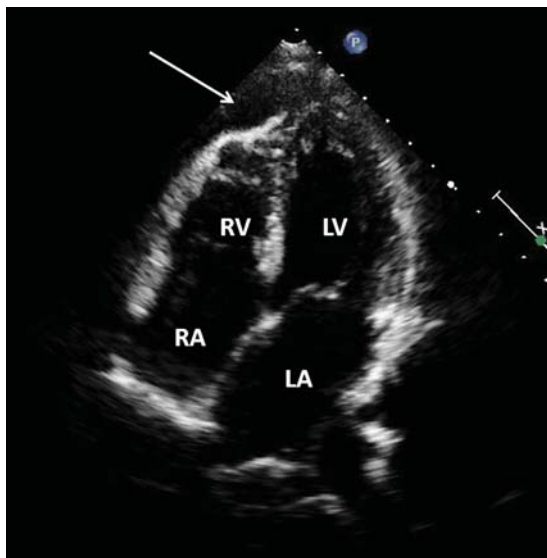


Figure 1 Echocardiographic image of a patient with pectus excavatum. The four-chamber view shows an echofree space at the apex consistent with the pericardial effusion (arrow) mostly seen around the right ventricular apex. RV, right ventricle; RA, right atrium; LV, left ventricle; LA, left atrium.

was only described by CMR ($P = 0.06$). The RV contractility was mildly reduced in two patients by echocardiography and in eight patients by CMR: seven patients with a borderline RV ejection fraction of 46–52% and only one patient with a significant reduction of RV ejection fraction of <45%. Minor criteria described for ARVC were present in 7 patients by echocardiography and in 10 patients by CMR (56%) ($P = 0.45$). Overall, there were frequent discrepancies in these patients with significantly diminished echo quality (see Table 5) but no significant differences between CMR and echocardiography.

Discussion

In patients with PEX an accurate cardiac assessment is important as many of them are referred to cardiologists either because of having cardiac symptoms, suspected congenital heart disease, and/or having an enlarged heart contour on chest X-ray.¹⁷ In the current study, we found frequent pericardial effusion and cardiac displacement with consecutive RV morphological anomalies by both, echocardiography and CMR. In some of these RV findings resembling ARVC, however, no patient fulfilled the criteria for ARVC.¹⁶ Pericardial effusion was common but never of clinical relevance, which may render further extensive evaluation of pericardial effusion in these patients unnecessarily. Discrepant findings between echocardiography and CMR were frequent (probably due to the diminished echocardiographic quality in many of these patients due to the PEX) but not significant. No method was more sensitive than the other.

Distortion of right-sided cavities resembling arrhythmogenic right ventricular cardiomyopathy

In PEX the right heart cavities are often impressed, distorted, and/or displaced between the spine and the sternum. This may potentially cause abnormal RV anatomy and function. Patients with PEX echocardiographic findings similar to ARVC were first reported by Mocchegiani et al.,¹⁸ who assessed RV morphology and function using echocardiography and chest radiography. They described localized wall anomalies of the RV wall, changes in the RV apex and structural changes of the moderator band resembling ARVC. Our data confirm these observations, as in our study anomalies similar to ARVC occurred in more than half of the patients, shown by both modalities, echocardiography and CMR. These morphological abnormalities may be related to the mechanical compression and/or distortion of the right atrium and the RV between the sternum and the vertebral column. This distortion can cause tractions on the RV wall resulting in wall thinning; localized wall anomalies were observed mainly between the RV

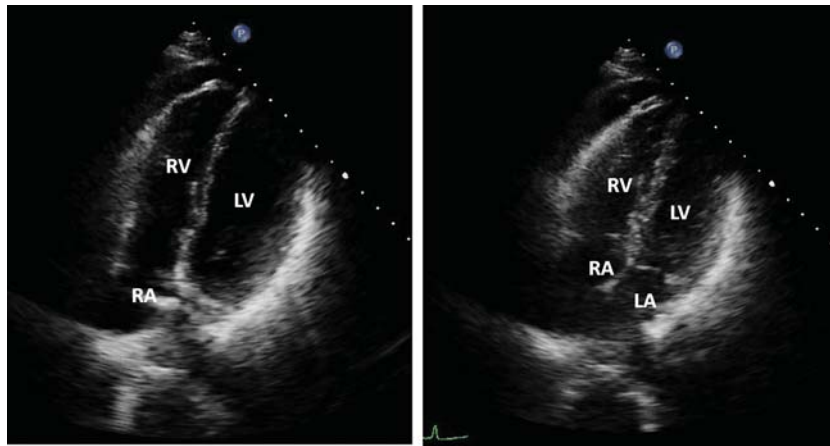


Figure 2 Apical four-chamber view by echocardiography of a patient with pectus excavatum demonstrating diminished echocardiographic quality in the presence of cardiac displacement. Pericardial effusion (arrow) can be recognized at end diastole (left side) and end systole (right side). RV, right ventricle; RA, right atrium; LV, left ventricle; LA, left atrium.

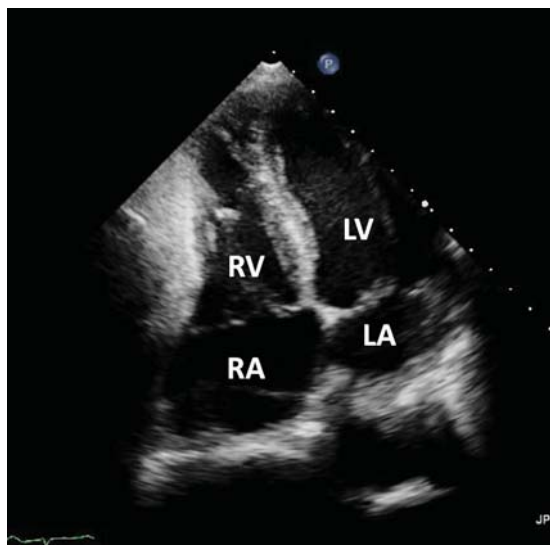


Figure 3 Typical echocardiographic image in pectus excavatum at end diastole (left) and end systole (right) with visible localized wall anomalies (arrows) at end diastole and a mildly dilated right ventricle. RV, right ventricle; RA, right atrium; LV, left ventricle; LA, left atrium.

trabeculations, which appeared to exert more traction than normal on the RV free wall (Supplementary data online, *Movies S1–S3*). Beside these morphological changes, in patients with PEX symptoms similar as in ARVC may occur.¹⁹ Therefore, careful exclusion of ARVC as potential fatal disease, with sudden cardiac death occurring in 10% of the patients, is important.²⁰

Pericardial effusion

A remarkable finding of our study was the high prevalence of pericardial effusion (example Supplementary data online, *Movie S4*).

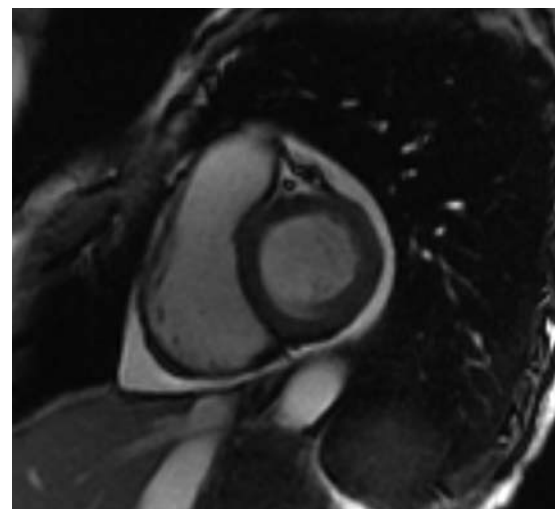


Figure 4 Steady-state free precession short-axis view by cardiac magnetic resonance imaging demonstrating a bright layer of fluid (arrows) around the posterior wall of the left ventricle, which represents the pericardial effusion.

Several other studies described PEX findings by CMR, echocardiography or computed tomography, but pericardial effusion has never been described before in these patients. Interestingly, one case report on a patient with PEX shows an image with an obvious pericardial effusion, without commenting this finding.²¹ It is unclear how pericardial effusion can be caused by PEX; we postulate a mechanical irritation of the pericardium, effusion *ex vacuo*, and/or disturbed fluid absorption as possible causes. In a single case report of a 15-year-old girl with PEX and pericardial effusion, the anterior posterior compression of the heart was assumed to be the causing mechanism.²¹ In a previous report, a review of

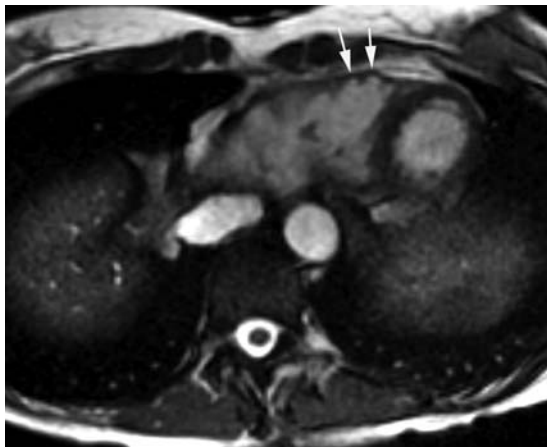


Figure 5 Steady-state free precession axial image by cardiac magnetic resonance imaging showing microaneurysms of the right ventricular anterior wall.

Table 5 Comparison of findings of echocardiography and CMR in 18 patients with severe PEX

	Only by echocardiography	Only by CMR
Localized RV anomalies	2 patients	3 patients
Pericardial effusion		4 patients
Diminished RV function	1 patient	8 patients
Any sign of ARVC	1 patient	6 patients

three large series of patients with pericardial effusions revealed that the most common causes of pericardial effusion are infection, cancer, or idiopathic cause,²² but PEX was not mentioned. Even though pericardial effusion was a frequent finding in our patients, no pericardial effusion was haemodynamically significant. Therefore, by knowing that PEX is frequently associated with insignificant pericardial effusion, we suggest that in these patients investigations may be limited to a clinical examination and thyroid function testing.

Associated congenital heart disease

In a group of 79 patients with PEX consecutively seen at our centre (unpublished data), any congenital heart disease (CHD) was present in 15 patients (19%), including a secundum atrial septal defect in six patients (8%), a ventricular septal defect in five patients (6%) and a partial atrioventricular septal defect in four patients (5%). Among the patients included in this study, only one had a haemodynamically insignificant tiny atrial septal defect. Therefore, CHD seems to be more common in patients with PEX than in the normal population, and exact cardiac assessment in these patients is essential.

Comparison between echocardiography and CMR findings

The differences observed between echocardiography and CMR regarding quantification of the right-sided cardiac structures and assessment of morphological anomalies reflect the characteristics of both imaging modalities. In PEX the acoustic window of echocardiography is frequently severely impaired by the chest deformity, what renders accurate RV assessment even more difficult. Localized wall anomalies of the anterior RV wall, which lies directly behind the deformed chest wall, may be hard to recognize by echocardiography. In contrast, image quality of CMR is totally independent from chest wall abnormalities, and its diagnostic capabilities remain unchanged. Moreover CMR is considered the gold standard for quantification of RV size and function, and remains the only non-invasive imaging modality able to provide this information. Three-dimensional (3D) echocardiography represents a valuable alternative for assessing ventricular volumes; however, patients with PEX may not be good candidates to be examined by 3D echocardiography, due to the limited echocardiography window.^{19,23,24}

Measurements of RVOT size cannot be directly compared, when measured with two different techniques. In CMR and in echocardiography, measurements were taken in different projections. In CMR, RVOT measurements were made on a right anterior oblique view tailored through the RVOT, and diameters were measured below the pulmonary valve. By echocardiography, RVOT diameter was measured above the aortic valve in a parasternal short-axis view as described previously. Thus, the observed differences may mainly be due to a methodological difference.

Pericardial effusion was detected by echocardiography and by CMR in the same 6 of 10 patients. In one patient a very small layer of pericardial effusion was described by echocardiography, but interpreted as epicardial fat at CMR; in four patients, small pericardial effusions were only described by CMR. Generally, CMR has been described to be more sensitive than echocardiography for detecting small pericardial effusions; CMR allows complete visualization of the pericardial sac independently from any thoracic deformities.²⁵

Limitations

This study is limited by the small number of patients included. Nevertheless, to our knowledge, this is the largest study assessing cardiac findings in consecutive PEX patients referred for echocardiography and comparing these findings to CMR. As these patients were referred for echocardiography, many of them had mild cardiac symptoms, however, this is not surprising as they all had rather significant PEX which is known to cause dyspnoea on exertion or an increased incidence of palpitations. However, a referral bias cannot be excluded.

The echocardiographic and CMR studies were not performed on the same day, but within 30 days of each other. We do not believe that this may have affected our findings.

We describe a high prevalence of morphological findings similar to ARVC. The presence of this cardiomyopathy seems improbable, as other criteria such as syncope, ECG changes or ventricular arrhythmias were lacking. However, genetic screening of the

patients was not performed, so that the disease cannot be ruled out definitely. Nevertheless we believe that the concomitant occurrence of significant PEX and ARVC in these patients is unlikely. These RV abnormalities are most likely caused by cardiac translation to the left and RV impression. This is important to know for any echocardiographer to prevent unnecessary cardiac screening.

Contrast imaging by CMR was not performed in this study. Post-contrast imaging may provide additional information about the presence of fibrosis; however correct interpretation of the images is difficult in the thinned RV free wall. Moreover, either the presence of fatty acid infiltration, or fibrosis is a diagnostic criterion in the new modified Task Force criteria for ARVC.¹⁶ Therefore, we do not believe to have misrecognized any case of ARVC solely because we did not perform post-gadolinium imaging; additional criteria need to be present for diagnosing ARVC.

None of these patients had previous radiotherapy or myocardial infarction as a cause of pericardial effusion. Hypothyroidism was excluded in all patients and in none of the patients there were signs of viral infection, tuberculosis, collagen vascular disease, or malignancy. Of course this is not a definite proof that pericardial is only due to PEX.

Conclusion

This prospective study demonstrates that in patients with PEX cardiac anomalies can be frequently detected by both echocardiography and CMR. Findings include pericardial effusion and morphological anomalies of the right ventricle sometimes resembling ARVC. We postulate that most of these anomalies are caused by the mechanical distortion of the heart. Usually, by combining clinical history, ECG criteria and findings at cardiac imaging, ARVC can be excluded. Pericardial effusion is mainly clinically irrelevant and may not need further evaluation.

Supplementary data

Supplementary data are available at *European Heart Journal—Cardiovascular Imaging* online.

Conflict of interest: none declared.

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