Two-dimensional strain as a marker of subclinical anterior ischaemia in anomaly of left coronary artery arising from pulmonary artery

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A 13-year-old boy was admitted to our department after an out-of-hospital cardiac arrest during physical exertion. Transitory ST-segment elevation in the anterior chest leads was noted after defibrillation. At 48 h, initial evaluation was performed. Twelve-lead EKG and telemetry were normal. Transthoracic echocardiography showed normal left ventricle (LV) size and global function. Segmental two-dimensional (2D) longitudinal strain of the anterior wall was significantly decreased when compared with the other segments, and was associated with post-systolic shortening. Coronary angiography and 64-slice computed tomography revealed an anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA). ALCAPA is a rare congenital malformation. The usual clinical course is a severe left-sided heart failure and mitral valve insufficiency presenting during the first months of life. In some cases, collateral blood supply from the right coronary artery is sufficient and symptoms may be subtle or even absent. However, ventricular arrhythmias or sudden cardiac death during exercise may be the first clinical presentation in patients with ALCAPA. Indirect evidence suggests that myocardial ischaemia is the underlying aetiology of cardiac ventricular ischaemia in patients with ALCAPA. Post-systolic shortening and altered longitudinal strain have recently been described as potential useful markers of ischaemic dysfunction in patients with ischaemic heart disease. In this case report, we demonstrate the usefulness of 2D strain as a non-invasive tool to assess subclinical myocardial ischaemia in patients with an ALCAPA. This provides further supportive evidence for the role of cardiac ischaemia in aetiology of ventricular arrhythmia in this rare condition.

KEYWORDS
2D strain; Ischaemia; Sudden death; ALCAPA

A 13-year-old boy was admitted to our department after an out-of-hospital cardiac arrest during physical exertion. Transitory ST-segment elevation in the anterior chest leads was noted after defibrillation (Figure 1). At 48 h, initial evaluation was performed. Twelve-lead EKG and telemetry were normal. Transthoracic echocardiography showed normal left ventricle size and global function (see Supplementary data online, AVI files). Segmental two-dimensional (2D) longitudinal strain of the anterior wall was significantly decreased when compared with the other segments (Figure 2), and was associated with post-systolic shortening (Figure 3). Coronary angiography and 64-slice computed tomography revealed an anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) (Figure 4).

ALCAPA is a rare congenital malformation. The usual clinical course is a severe left-sided heart failure during the first months of life. In some cases, collateral blood supply from the right coronary artery is sufficient and symptoms may be subtle or even absent. However, ventricular arrhythmias or sudden cardiac death during exercise may be the first clinical presentation. Indirect evidence suggests that myocardial ischaemia is the underlying aetiology of cardiac ventricular ischaemia in patients with ALCAPA.4 Post-systolic shortening and altered longitudinal strain have recently been described as potential useful markers of ischaemic dysfunction in patients with ischaemic heart disease.4,5 In this case report, we demonstrate the usefulness of 2D strain as a non-invasive tool to assess subclinical myocardial ischaemia in patients with an ALCAPA. This provides further supportive evidence for the role of cardiac ischaemia in aetiology of ventricular arrhythmia in this rare condition.

Supplementary data
Supplementary data are available at European Journal of Echocardiography online.
Figure 1  (A) Initial EKG showing ventricular fibrillation and reduction after 100 J external electric shock. (B) EKG after shock. (C) EKG 5 min latter showing normalization of ST-segment.

Figure 2  Bull’s eye map of peak systolic strain showing abnormal contraction in the anterior, anteroseptal, and anterolateral walls.
Figure 3  (A) 2D strain segmental analysis in apical two-chamber view. Medial part of the anterior and the inferior wall are marked by a yellow and a blue cross, respectively. Strain analysis of these segments is represented in the graph in (B). (B) Post-systolic shortening of the medial segment of the anterior wall (yellow line, white arrow) and normal contraction of the medial part of the inferior wall (blue line). Aortic valve closure is indicated by the dotted vertical green line.

Figure 4  (A) Coronary CT-angiography showing normal course of a very large right coronary artery. (B) Anomalous origin of the left main coronary artery from the pulmonary artery. (C) Aortic angiography showing a very large right coronary artery and absence of left main coronary artery. (D) Selective angiography of right coronary artery with collateral filling of the left coronary vascular territory, which connects directly to the pulmonary trunk.
References