A rare coronary anomaly unmasked by ST abnormalities on 24-h Holter: a case report

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INTRODUCTION

Abnormalities of coronary sinus are rare congenital diseases with potential for sudden cardiac death.1 Coronary artery anomalies occur in approximately 1.3% of patients undergoing coronary angiography and 0.3% of an autopsy series.2 The diagnosis requires a high clinical suspicion since its presentation is quite unspecific and variable. In young adult athletes, coronary anomaly is an important cause of sudden death during intense exercise.3 There is a broad diversity of anatomical abnormalities and can be associated with structural cardiac malformations, leading to sudden death such as coronary artery disease.

CASE PRESENTATION

A 21-year-old female with no pathological medical history was referred to the emergency department with significant dyspnea during physical exertion during the last 6 months. The symptoms became more intense (walking few meters) in the last month. On examination, the patient had normal jugular venous pressure and clear lungs. Her blood pressure was 128/66 mmHg, her heart rate was 72 beats per minute and respiratory rate was 24 breaths per minute. Her electrocardiogram at rest showed normal sinus rhythm at 72 beats per minute, with no specific ST-segment or T-wave abnormalities. Chest X-ray was normal and two-dimensional echocardiography (2D-eco) showed preserved ejection fraction of the left ventricle (0.71), mild mitral insufficiency and no wall-motion or diastolic abnormalities. On treadmill stress test, the patient did not reach 85% of her maximum predicted heart rate and was negative for myocardial ischemia. The results showed low tolerance and functional capacity in the treadmill stress test.

A 24-h Holter was requested to evaluate the presence of significant dyspnea during physical exertion during the last 6 months. Her rest electrocardiogram did not show specific ST segment or T wave abnormalities and the two-dimensional echocardiogram was normal. Because she reported palpitation a 24-h Holter was applied and it showed ST segment depression reaching up to 2.5 mm. Considering the symptoms of dyspnea in a young patient with not apparent anatomical cause by two-dimensional echocardiogram, a coronary computed tomography angiography was performed. It demonstrated anomalous origin of right coronary with trajectory between aorta and pulmonary artery and no coronary obstruction in coronary artery. After diagnosis, the patient was submitted to surgical reimplantation of the ostium of the right coronary. During the in-hospital post-operative stay, the patient had full recovery and no complications. She was discharged briefly and returned asymptomatic one month after the procedure.
arrhythmias and ST segment during daily activities. It showed ST segment depression reaching up to 2.5 mm during exertion and 29 isolated ectopic supraventricular beats (Fig. 2). Considering symptoms of dyspnea without apparent anatomical cause by 2D-echo and normal diastolic parameters, a coronary computed tomography angiography (CCTA) was performed. It showed zero calcium score, no coronary obstruction in coronary artery and anomalous origin of right coronary (RC) with trajectory between aorta and pulmonary artery were noted (Figs. 3 and 4). After diagnosis, the patient was submitted to surgical reimplantation of the ostium of the RC artery. During the in-hospital post-operative stay, the patient had no complications and full recovery. The patient was discharged in the third postoperative period. The patient returned to ambulatory unit 1 month after the procedure, presenting asymptomatic.

**DISCUSSION**

We describe a rare case of coronary anomaly that was unmasked after findings of ST segment on 24-h Holter leading to the indication of CCTA focusing on suspicion of coronary artery as the cause of symptoms. The clinical suspicion of the presence of coronary artery anomaly based only on clinical findings is a great challenge. Symptoms tend to be later, occurring mainly earlier in adult life, when heavy exercise can cause intolerance in previously compensated patients. Most patients remain asymptomatic for long lifetime; however, they may die suddenly. Most of these patients may experience symptoms of chest pain and syncope, which sometimes precede a fatal event.3

The diagnosis of these coronary abnormalities may not be established by changes in treadmill stress test as described in this patient. Exams, like echo-stress or nuclear imaging stress test may detect ischemia in cases of negative treadmill test but the best non-invasive exam to detect coronary anomalies is the CCTA. The angiography by CT can show anatomical details of coronary ostium and its relation with other structures such as pulmonary artery. However, in general, the CCTA is performed after a consistent suspicion based on clinical data and common exams. Among these exams commonly performed in clinical practice, 24-h Holter
monitoring can be a source of valuable information as reported in the current case.

After the initial suspicion, a CCTA was performed and the diagnosis was confirmed. After the definition of this diagnosis, the practice of sports should be prohibited and surgical correction, when feasible, can repair the coronary flow. The definitive treatment of anomalous coronary origin must be guided by anatomical characteristics of coronary ostium. It can be performed surgical ostium reimplantation for suitable cases similar to a myocardium revascularization surgery.

In summary, we describe a rare case of right coronary anomaly whose persistence of the dyspnea and lowered ST segment on 24-h Holter led to clinical suspicion of coronary obstruction. A CCTA showed a rare presentation of coronary anomaly and surgical treatment was indicated. After the procedure, the patient had improvement of the symptoms and early hospital discharge, brief outpatient return and no cardiovascular symptoms.

**REFERENCES**