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CLINICAL CASE

Coronary arterio-venous fistula associated acute coronary syndrome: A case-report and review of literature

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KEYWORDS
Chest pain;
Acute coronary syndrome;
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Abstract A 56-year-old woman with no medical history presented to the emergency department complaining of oppressive chest pain of three hours of duration triggered by walking. An electrocardiogram (ECG) performed at hospital admission showed ST-segment depression and T-wave inversion in left precordial and inferior leads. Serum troponin I levels were above the normal values. Non ST-segment elevation acute coronary syndrome (NSTEACS) was diagnosed and an invasive approach with a percutaneous coronary intervention was performed. No obstructive coronary lesions were observed. However a tortuous coronary fistula emerging from the left-anterior descending coronary artery to the pulmonary artery was observed. Coronary arterio-venous fistulas are present in 0.002% of the general population and in 0.25% of patients undergoing cardiac catheterization for any cause. Most of them are asymptomatic. Ischemic symptoms may develop but are infrequent in patients with no atherosclerotic disease.

PALABRAS CLAVE
Dolor torácico;
Síndrome coronario agudo;
Fistula coronaria arterio-venosa

Síndrome coronario agudo asociado a fístula coronaria arterio-venosa: reporte de un caso y revisión de literatura

Resumen Un hombre de 56 años sin antecedentes médicos se presentó en el departamento de urgencias quejándose de dolor torácico opresivo de tres horas de duración desencadenado al caminar. El electrocardiograma (ECG) tomado al ingreso mostró depresión del segmento ST e inversión de las ondas T en derivaciones precordiales izquierdas e inferiores. Los niveles

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de troponina I sérica se encontraron arriba de los valores normales. Se diagnosticó síndrome coronario agudo sin elevación del segmento ST (SCACEST) y se realizó un abordaje invasivo mediante intervención coronaria percutánea. No se observaron lesiones coronarias obstructivas. Sin embargo, se observó una fístula coronaria tortuosa emergiendo de la arteria coronaria descendente anterior hacia la arteria pulmonar. Las fístulas arterio-venosas coronarias se encuentran presentes en 0.002% de la población general y en 0.25% de los paciente sometidos a cateterismo cardíaco por cualquier causa. Muchos de ellos son asintomáticos. Síntomas isquémicos se pueden desarrollar, pero son infrecuentes en pacientes sin enfermedad aterosclerosa.

Introduction

Coronary arterio-venous fistulas (CAVF) are rare major vascular malformations in which there is an abnormal connection between a coronary artery and any of the four chambers of the heart or any of the great vessels (superior vena cava, pulmonary artery, pulmonary veins or coronary sinus), by-passing the coronary bed.[1–5,7–13]

It is estimated that CAVF are present in 0.002% of the general population, represent 0.4% of all cardiac malformations and are visualized in nearly 0.25% of patients undergoing catheterization.[1,12] At least 75% of coronary artery fistulas found incidentally are small and clinically silent. However the true incidence of coronary artery fistulas is highly speculative since many may be small and approximately 50% remain asymptomatic, or are only detected incidentally with imaging for another indication.[1,2,6,7,9–12]

CAVF was first described by Krause in 1865 and the first successful surgical closure was reported by Bjork and Crafoord in 1947[1–7] in a patient with a preoperative diagnosis of patent ductus arteriosus,[2] but their diagnostic triad was not described until 1978 by Haller and Little, who characterized these lesions by the presence of an abnormal continuous murmur similar to that of a patent ductus arteriosus, a left-to-right shunt, and a large coronary artery with evidence of fistula on angiography.[3–5]

CAVF may be congenital or acquired.[1–3,5,10,12] The congenital form is by far the most frequent and represents an arterial anomaly of termination (it terminates into an abnormal structure). It can be found in any age group and has no predilection among both sexes. Acquired CAVF are extremely rare and usually iatrogenic, posttraumatic or caused by Takayasu arteritis or chest irradiation.[5,12]

Congenital CAVF may arise due to persistence of sinusoidal connections between the lumens of the primitive tubular heart that supply myocardial blood flow in the early embryologic period.[2,10] Spontaneous closure of the fistula secondary to spontaneous thrombosis has been reported, although it is very uncommon (1–2% of cases).[1,2,4,5,7]

Two major groups have been identified; solitary and multiple CAVF.[7] Single fistulas are more common, ranging from 74 to 90%. Multiple fistulae are present in up to 16%, and fistulas originated from both coronaries in 5%.[5,12] The solitary form is that observed in acquired CAVF.[7] 3% of cases are associated with an absence of the contralateral coronary artery.[12] Some CAVF might disappear spontaneously during childhood,[1,2,12] specially with small or medium size fistulas,[11] although this is rare.[4]

Approximately 10–30% of patients with a CAVF also have another congenital cardiovascular anomaly. The most commonly seen defects include tetralogy of Fallot, patent ductus arteriosus, and atrial septal defect.[5,6,11,12]

CAVF are classified according to the chamber or vessel to which it drains: type 1, draining to the right atrium; type 2, draining to the right ventricle; type 3, draining to the pulmonary artery; type 4, draining to the left atrium; and type 5, draining to the left ventricle.[6] Drainage into the left-sided chambers is less frequent. Fistulous drainage occurs into the right ventricle in 40%, right atrium in 26%, pulmonary artery in 17%, left ventricle in 3%, coronary sinus in 7%, and superior vena cava in 1%.[1,3,5,6,10]

Case report

A 56-year-old woman with no medical history presented to the emergency department complaining of oppressive chest pain and dyspnea three hours lasting, triggered by walking.

An electrocardiogram (ECG) performed at hospital admission showed sinus rhythm; and 0.1 mm ST-segment depression and T-wave inversion in precordial leads from V-3 to V-6 and in the inferior leads DII, DIII and aVF (Fig. 1). Blood samples were obtained and serum troponin I levels were 1.5 ng/ml which was above the 99th percentile upper reference limit. No other remarkable findings were reported.

Non ST-segment elevation acute coronary syndrome (NSTEACS) was diagnosed and an invasive approach with a percutaneous coronary intervention was performed at the second hour from hospital admission. No obstructive coronary lesions were observed. However a tortuous coronary fistula with multiple emerging branches from the left-anterior descending coronary artery to the pulmonary artery was visualized (Fig. 2A and B).

An interventional approach for the correction of the fistula was first intended, however given the complexity of the vascular malformation, a surgical approach was offered to the patient. For no medical reasons surgery was rejected by the patient and a medical treatment was implemented.