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Technical options for the treatment of anomalous origins of right or left coronary arteries associated with aortopulmonary windows

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The association of an aortopulmonary window (APW) and an anomalous origin of a coronary artery is an exceptional condition with pathophysiologic specificities that make its preoperative diagnosis difficult. We report 2 new cases of APW with anomalous origin of either the right coronary artery (RCA) or the left coronary artery (LCA) and discuss surgical options for their repair.

CLINICAL SUMMARY

A 8-year-old girl (17 kg) was admitted for an APW revealed by a systolic murmur. Electrocardiogram showed a sinus rhythm and no ischemic sign. The chest radiograph demonstrated increased pulmonary vascularization, and the cardiothoracic ratio was 0.7. The echocardiography demonstrated a left ventricular dilatation, low-grade mitral and aortic regurgitations, a large APW, and a severe pulmonary hypertension. The origin of the LCA was identified in a normal position, but the origin of the RCA was not seen. Under normothermic cardiopulmonary bypass, after a vertical aortotomy, the left ostium was seen in the left sinus and the right ostium was located on the pulmonary side of the APW. The pulmonary trunk was transected, and the RCA was excised with a large button of pulmonary artery wall. This defect was repaired with an autologous pericardial patch. The RCA was reimplanted in the right aortic sinus, and the APW was closed with a polytetrafluoroethylene patch (Figure 1). The postoperative course was uneventful, without

ischemia on the electrocardiogram or on echocardiography. At 2 years, follow-up, the patient has a normal ventricular function with New York Heart Association class I.

A 7-month-old boy (7 kg) was admitted for an APW diagnosed on a systolic murmur and a mild dyspnea. Electrocardiogram showed no ischemic sign. On chest radiograph, the cardiothoracic ratio was 0.6 and an increased pulmonary vascularization was noted. Echocardiography demonstrated a left ventricular dilatation and a large APW with isosystemic pulmonary hypertension. No anomaly of coronary arteries was identified. Under normothermic cardiopulmonary bypass, a vertical aortotomy was performed and the LCA ostium was seen on the posterior wall of the pulmonary trunk. The repair consisted in a tunnel made of the equine pericardial patch used to close the APW, leaving the LCA ostium on the aortic side of this patch. A small polytetrafluoroethylene patch was used to close the aortotomy. The postoperative course was uneventful. The echocardiography performed the day after surgery revealed a left ventricular dysfunction and grade I mitral regurgitation. Both were regressive on echocardiography at discharge, 9 days after surgery. A postoperative computed tomography scan was performed to assess the tunnel patency (Figure 2). At 4 months follow-up, the patient is asymptomatic.

DISCUSSION

The association of an APW and anomalous origin of a coronary artery is an exceptional condition. It was first described by Stopfkuchen and colleagues¹ for the RCA, by Bournon and colleagues² for the LCA, and by Chopra and colleagues³ for the circumflex artery. To date, 11 RCAs, 2 LCAs, and 2 circumflex anomalous coronary arteries with APW have been reported in the literature.

Diagnosis is difficult with the lack of myocardial ischemic symptoms because of the peculiar pathophysiology encountered in this association: The abnormal coronary vessel originating from the pulmonary artery is perfused by a well-

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FIGURE 1. Surgical procedure performed for the reimplantation of the RCA and the closure of the APW.

oxygenated high pressure blood flow through the APW. Greenway and colleagues⁴ highlight the importance of echographic investigation of the coronary arteries in such cases. They report that extensive collateral flow within the myocardium represents a major clue. This indirect sign leads to a focus on the coronary ostia, which may be difficult to identify because of turbulence induced by the APW, and to complete investigations by angiography or computed tomography scan examinations. Finally, an anomalous origin of a coronary artery should be identified by intraoperative investigations, even if such an anomaly has already been ignored during APW closure.³

Ligation of the anomalous coronary artery or absence of treatment leads to myocardial ischemia after closure of the APW. Coronary artery bypass and proximal ligation have been used to treat ischemia in a teenager after closure of the APW in infancy³ but is not recommended for initial repair. Two types of procedures are possible: reimplantation or rerouting. Most authors have chosen a rerouting procedure, taking advantage of the APW. This technique is simple and preserves the native implantation of the coronary artery, avoiding the risk of kinking or stretching after transfer. Reimplantation can be a useful method, especially for the RCA,⁵ as in our first case, where rerouting by a patch may create a narrowed tunnel to the coronary artery.

CONCLUSIONS

Both methods are eligible for the repair of this rare anomaly, and the decision is based on the relationship between the anomalous coronary ostium and the APW.

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FIGURE 2. Postoperative computed tomography scan. LCA (black arrow); rerouting patch closing the APW (white arrow).