Case Report

Myocardial perfusion SPECT in a case of retropulmonary looping of left coronary artery in a baby after arterial switch surgery

Subramanyam Padma, Palaniswamy Shanmuga Sundaram

Department of Nuclear Medicine and PET CT, Amrita Institute of Medical Sciences, Amrita Vishwa Vidyapeetham University, Cochin, Kerala, India

ABSTRACT

Pediatric myocardial perfusion imaging (MPI) is not a routine investigation in an Indian setting due to under referrals and logistic problems. However, MPI is a frequently performed and established modality of investigation in adults for the identification of myocardial ischemia and viability. We report myocardial perfusion scintigraphy in a case of retropulmonary looping of left coronary artery in a baby after arterial switch surgery. Adenosine stress MPI revealed a large infarct involving anterior segment with moderate reversible ischemia of the lateral left ventricular segment. Coronary angiogram later confirmed left main coronary artery ostial occlusion with retrograde collateral supply from dilated right coronary artery.

Keywords: Arterial switch operation, myocardial perfusion imaging, retropulmonary looping of coronaries, transposition of great vessels, Taussig–Bing anomaly

INTRODUCTION

Asymptomatic patients post arterial switch (ASO) surgeries are at low risk for myocardial ischemia, and much depends on the type of coronary anomalies and the method of transfer of the coronary arteries during surgery. However certain coronary patterns in transposition of great vessels like a retropulmonary looping of left coronary artery (LCA) or an intramural coronary artery predict increased morbidity and mortality. Pediatric myocardial perfusion imaging (MPI) is an established investigation in the evaluation of ischemia and myocardial viability. It is said that ASO surgeries are at a low risk for myocardial ischemia, and much depends on the type of coronary anomalies and the method of transfer of thecoronary arteries during surgery. However, certain coronary patterns in transposition of great vessels such as aretropulmonary looping of left coronary artery or an intramural coronary artery predict increased morbidity and mortality.

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CASE REPORT

The patient was a first-born male child from a nonconsanguineous marriage. He was diagnosed with Taussig–Bing anomaly, large subpulmonic ventricular septal defect (VSD), severe coarctation with long-segment hypoplastic arch, retropulmonary looping of LCA, and a large patent ductus arteriosus at birth. The patient underwent arterial switch operation, bovine pericardial patch closure of VSD, resection of coarctation and reconstruction with tissue-to-tissue anastamosis posteriorly, and bovine pericardial patch augmentation anteriorly on 5th day of life. He presented with breathlessness at 5 months follow-up. Echocardiogram revealed no residual VSD, no regional wall motion abnormalities, and showed no evidence of any left ventricular outflow tract obstruction. Adequate biventricular function was confirmed, normal diaphragmatic movements were seen, and no pericardial effusion was noted.

In view of persistent breathlessness (functional classification II), the patient was referred for MPI with single-photon emission computed tomography (SPECT) to rule out any myocardial ischemia. Pharmacological same day stress–rest-gated MPI was performed under the supervision of a pediatric cardiologist using intravenous infusion of adenosine at a dose of 140 μm/kg per min for 6 min. At 3rd min of the infusion 2 mCi (millicurie) of ^{99m}Technetium sesta MIBI (^{99m}Tc-MIBI) was injected

Address for correspondence:
Dr. Subramanyam Padma, Department of Nuclear Medicine and PET CT, Amrita Vishwa Vidyapeetham University, Cochin - 680 2041, Kerala, India. E-mail: padmas@aims.amrita.edu

intravenously. Delayed stress-gated SPECT images were acquired after 1 h using high-resolution collimators on a dual-head variable angle siemens E Cam Gamma Camera.

Six millicuries of ^{99m}Tc MIBI was injected 3 h later at rest. Delayed rest-gated SPECT images were acquired. Motionless stress and rest acquisition were performed once the baby was sedated with intravenous midazolam at a dose of 0.1-0.2 mg/kg body weight. SPECT images of myocardium were acquired at 76 degrees cardiac angle using a 180-degree rotation from the right anterior oblique to the left posterior oblique projection. High-resolution collimators were used. Images were acquired in Tc window centered at 140 keV photopeak with a window of 20%. Images were acquired in a step and shoot mode, 64 × 64 matrix, for 74 frames at 25 s/frame with a zoom of 2.0.

Images were checked for motion during acquisition and were then processed as per the company provided reconstruction software into short axis, horizontal long axis, and vertical long axis. All images were interpreted visually and quantitatively using Emory cardiac Toolbox (ECTbox, Emory University, Atlanta, GA, USA).

Post–stress-gated SPECT images [Figure 1a] showed a fixed perfusion defect involving entire anterior segment (small arrow) and reversible perfusion defect in lateral segment (bold arrow) indicating a large LAD territory infarct with moderate ischemia in lateral segment of left ventricle.

The patient was further taken up for coronary angiogram to confirm the above findings and also to see the coronary artery relationship with right ventricular outflow tract (RVOT). Baseline pressures were recorded. Right coronary artery (RCA)-guiding catheter and terumo combination was used to enter into the RVOT and angiogram was done. A 0.5 Fr pigtail/terumo combination was used to enter the aortic root and angiogram was done simultaneously. A 14 × 4 cordis balloon was inflated across RVOT via 8F long sheath over V-18 wire to observe the relationship between the coronary artery and the RVOT. Aortis root angiogram showed a dilated RCA with delayed and retrograde filling of LCA through collaterals [Figure 1b]. The child had severe LCA origin stenosis and retrograde filling of LCA through collaterals. He had right ventricular (RV) pressures slightly more than half systemic, hence RVOT stenting was not done. After balloon dilatation of RVOT with 14 × 4 cordis balloon, there was no significant fall in the RV pressures. He has been advised a close follow-up.

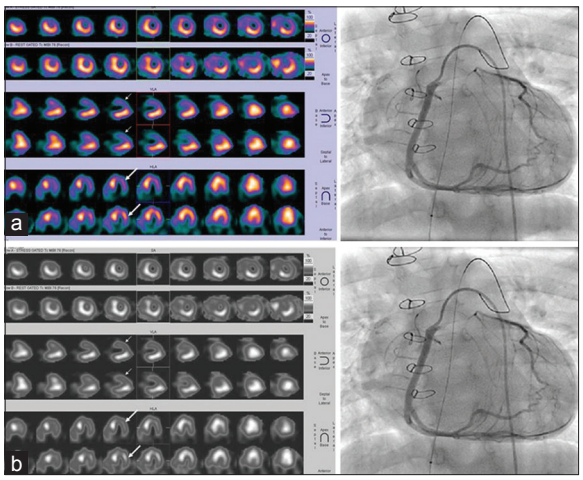

Figure 1: (a) Post–stress-gated SPECT images showed a fixed perfusion defect involving entire anterior segment (small arrow) and reversible perfusion defect in lateral segment (bold arrow) indicating a large LAD territory infarct with moderate reversible ischemia in lateral segment of left ventricle. (b) Coronary angiogram confirmed left main coronary artery ostial occlusion, with retrograde collateral supply from dilated right coronary artery

DISCUSSION

Taussig–Bing anomaly is a rare congenital heart malformation first described by Helen B Taussig and Richard J Bing.[1] It is the transposition of the aorta to the right ventricle and malpositions of the pulmonary artery with subpulmonary VSD.

Reports show an increased mortality and morbidity, especially in patients with additional anomalies such as transposition of great vessels such as retropulmonary looping of left coronary artery.[2] Pasquali *et al.*[3] reported that single coronary patterns, both of which loop around the great vessels, were associated with significant mortality [odds ratio (OR) 2.9, 95% confidence interval (CI) 1.3-6.8], whereas looping patterns that arose from two separate ostia were not (OR 1.2, 95% CI 0.8-1.9).

The current standard procedure includes mobilization and reimplantation of the coronary arteries into the neoaorta. The most common complication is enlargement of the neoaortic root. Late complications include kinking of the coronary artery or a failed coronary arterial anastomosis. No guidelines exist concerning the follow-up of children undergoing ASO. The outcome of such surgeries depends on the variations in coronary anatomy and their successful transfer. There are mixed opinions predicting increased morbidity and mortality in certain types of coronary anomalies such as retropulmonary looping of LCA or an intramural coronary artery. Our case highlights the early onset of MI in a patient with retropulmonary looping of LCA. It also highlights the relatively asymptomatic presentation of a large LAD infarct with reversible ischemia in LCx territory–supplied left ventricular segments within 5 months of surgery. Other causes implicated by Wernovsky *et al.* were based on postoperative kinking or stretching of these coronary arteries after their transfer.[4,5]

Pasquali *et al.*[3] analyzed the relationship between coronary artery variants and mortality after ASO in a meta-analysis of 9 case series. They reported a prevalence of coronary stenosis in approximately 5%-8%.[6,7] At times, even complete occlusion of a main coronary goes unnoticed as it may remain completely asymptomatic but it has been associated with sudden death. MPI imaging has a high negative predictive value, 98%. Selective coronary angiography has been recommended after ASO; however, there is no consensus as to when it should be performed.

Reddy *et al.*[8] have recently published their data on adenosine SPECT MPI in 10 asymptomatic children post-ASO. They conclude that postadenosine stress MPI is safe and feasible in patients of ASO for transposition of great arteries.

CONCLUSION

This case report highlights importance of identifying coronary artery stenosis in the perioperative period of congenital heart disease patients, which may go unnoticed in the absence of high degree of clinical suspicion. It also reiterates that although post-ASO surgery patients are considered as at a low risk for ischemia, there is increased mortality and morbidity, especially in patients with additional anomalies such as transposition of great vessels such as retropulmonary looping of left coronary artery.

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