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Original Article / Özgün Makale

Dual source multidetector computed tomography coronary angiography in the surgical planning regarding the major coronary artery anomalies in children

Çocuklarda majör koroner arter anomalilerinin cerrahi planlamasında çift kaynaklı multi dedektör bilgisayarlı tomografi koroner anjiyografi

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Background: This study aims to demonstrate major coronary artery anomalies such as anomalous origin and coronary artery fistulas on dual-source multi detector computed tomography (MDCT) coronary angiography in children before surgical treatment.

Methods: Six children (3 boys, 3 girls; mean age 6 years; range 2 days to 15 years) referred for a MDCT coronary angiography for evaluation of coronary anomaly between January 2010 and June 2011 were included in this study. Electrocardiography (ECG)-gated dual-source MDCT coronary angiography was performed on all patients. No beta-blockers were given prior to the procedure. The mean heart rate was 98±20 bpm (range 128 to 75 bpm). In all patients, ostium, course, and end-distal portion of coronary artery were inspected for anomalies. Exact opening point was carefully assessed in patients with fistulas. Dual-source MDCT examination was performed with low kilovolt dose reduction algorithm (80 kV, range 150-350 mA) compatible with ALARA principles.

Results: Three anomalous origin (one ALCAPA, two anomalous left main coronary artery originated from right sinus of Valsalva) and three fistulas (two coronary-cameral fistula, one left main coronary artery -coronary sinus fistula) were clearly identified with dual-source MDCT angiography.

Conclusion: Dual source MDCT angiography is a noninvasive and effective method as an alternative to conventional coronary angiography for the preoperative assessment of major anomalous origin of coronary artery or coronary artery fistulas.

Key words: Children; coronary anomalies; dual-source computed tomography.

Amaç: Bu çalışmada, çocuklarda anormal orijin ve koroner arter fistülü gibi majör koroner anomalileri cerrahi tedavi öncesi çift kaynaklı multi dedektörlü bilgisayarlı tomografi (MDBT) koroner anjiyografi ile gösterildi.

Çalışma planı: Ocak 2010 - Haziran 2011 tarihleri arasında koroner anormalliğin MDBT koroner anjiyografi ile değerlendirilmesi amacıyla kliniğimize sevk edilen toplam altı hasta (3 erkek, 3 kız; ort. yaş 6 yıl; dağılım 2 gün-15 yıl) çalışmaya dahil edildi. Tüm hastalara elektrokardiyografi (EKG) tetiklemeli çift kaynaklı MDBT koroner anjiyografi yapıldı. İşlem öncesi beta-bloker uygulanmadı. Ortalama kalp hızı 98±20 atım/dk. (dağılım 128-75 atım/dk.) idi. Tüm hastalarda koroner arterlerin ostiumları, seyirleri ve distal uçları anomaliler açısından incelendi. Fistül saptanan olgularda açılma noktası dikkatlice değerlendirildi. Çift kaynaklı MDBT incelemelerinde düşük kilovolt tekniği ile ALARA ilkeleri ile uyumlu doz azaltıcı algoritma uygulandı (80 kV, dağılım 150-350 mA).

Bulgular: Üç anormal orijin (bir ALCAPA, iki sağ sinüs Valsalvadan çıkan anormal sol ana koroner arter) ve üç fistül (iki koroner-kameral fistül, bir sol ana koroner arter-koroner sinüs fistülü) çift kaynaklı MDBT ile ayrıntılı olarak gösterildi.

Sonuç: Çift kaynaklı MDBT, majör koroner arter anomalilerinin veya koroner arter fistüllerinin ameliyat öncesi değerlendirmesinde konvansiyonel koroner anjiyografiye alternatif noninvaziv etkili bir yöntemdir.

Anahtar sözcükler: Çocuklar, koroner anomaliler; çift kaynaklı bilgisayarlı tomografi.

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Early detection and evaluation of coronary artery anomalies are of significant importance before cardiac surgical planning in children as they serve as alternatives to conventional angiography.[1-3] Congenital coronary anomalies may be isolated or associated with other congenital heart diseases (CHD), including tetralogy of Fallot, transposition of the great arteries, and pulmonary atresia.[4] Conventional coronary angiography has been the gold standard technique for identification of coronary artery anomalies for several decades. However, besides its invasive nature, this technique has some major limitations for the visualization of ostial coronary anomalies.[5]

Various surgical strategies are available for patients with coronary artery abnormalities. Almost all surgeons prefer a primary complete repair procedure rather than a staged approach or interventional procedure. Preoperative detection of cardiac anatomic variations would allow them to decide on the most adequate and optional surgical procedure. At the preoperative evaluation, echocardiography is normally performed as a first-line examination technique in patients with CHD. In many cardiac centers, preoperative conventional coronary angiography is also routinely performed to evaluate the anatomy of coronary arteries, but the limitations and potential complications of cardiac catheterization are well known.[2,5-7]

Multidetector computed tomography (MDCT) angiography has emerged as a novel technique which has overcome most of the disadvantages of conventional coronary angiography. It provides precise, noninvasive evaluation of coronary artery anomalies with regard to origin, course, and termination.[1,7] With its high temporal and spatial resolution and advanced post-processing algorithms, MDCT creates high quality multiplanar images which aid in the evaluation of the coronary arteries along with the cardiac chambers, pulmonary arteries, and aorta. Dual source multislice CT with its higher temporal resolution (83 ms) allows for the evaluation of coronary arteries in everyday practice in newborns and children with high heart rates.[1,2,8]

PATIENTS AND METHODS

Between January 2010 and June 2011, a total of six selected children who underwent MDCT as a part of their preoperative evaluation were included in this study. Patient details are listed in Table 1.

Multidetector computed tomography protocol

Computed tomography coronary angiography (CTCA) was performed with dual source 64-channel MDCT (SOMATOM Definition, Siemens Medical Solutions, Erlangen, Germany). Coronary CT examinations were performed with a low kilovolt dose reduction algorithm (80 kV, range 150-350 mA based upon body weight) for all patients. A timing bolus was performed at 80 kVp and 10 mAs per rotation at the level of the left ventricle (LV). The maximum tube current pulse window was set for 35-75% of the cardiac cycle. The tube current was modulated down to 25% of the maximum mAs per rotation outside the pulse window.

Iodinated contrast material (320 mg of iodixanol per milliliter, Visipaque 320 mg/mL, GE Healthcare Inc., Princeton, New Jersey, USA) 1.5 ml per kilogram was

Table 1. Patient data

Case	Age	Weight (kg)	Diagnosis	Associated extracoronary findings	Causes of hospital admission	Treatment	HR during MDCT
1	2 months	4.5	ALCAPA	Heart failure and LV ischemia, PFO	Severe heart failure	Bypass surgery	126 bpm
2	11 years	38	LMCA originating from the RCA	No	Palpitation	Follow-up	80 bpm
3	7 months	14	CCF (LAD-RV)	RV dilatation	Heart murmur	Bypass surgery	126 bpm
4	10 years	18	CCF (RCA-RV)	Fontan procedure for Tricuspid atresia	Postoperative evaluation	Fallow-up	89 bpm
5	8 days	3.2	CCF (LMCA-CS)	VSD, PFO, PH	Heart murmur	Bypass surgery	136 bpm
6	15 years	45	LMCA from the right sinus of valsalva	Ischemia during exercise testing	Syncope during exercise	Bypass surgery	78 bpm

MDCT: Multidedector CT; HR: Heart rate; ALCAPA: Anomalous left coronary artery from the pulmonary artery; LV: Left ventricle; PFO: Patent foramen ovale; LMCA: Left main coronary artery; RCA: Right coronary artery; CCF: Coronary cameral fistula; LAD: Left anterior descending; RV: Right ventricle; CS: Coronary sinus; VSD: Ventricular septal defect; PH: Pulmonary hypertension.

injected into the right antecubital vein at a flow rate of 0.8-3 mL/seconds. This was followed by a saline chaser, which was injected at the same flow rate. A bolus-tracking technique was applied to synchronize the data acquisition with the arrival of contrast material in the coronary arteries.

Case 1– *Anomalous left coronary artery from the pulmonary artery (ALCAPA) syndrome:* A two-month-old girl was admitted to our hospital with severe dyspnea. Physical examination revealed a grade 2/4 systolic murmur, and cardiomegaly was noted on her chest X-ray. Electrocardiography (ECG) showed deep Q waves in leads I, aVL, and V4-V6 derivations, and echocardiography revealed a dilated and hypokinetic LV with an end-diastolic diameter of 36 mm and an ejection fraction of 18%. The left main coronary artery (LMCA) was not visible. There was also dilatation of the right coronary artery (RCA) and diastolic flow in the pulmonary artery (PA). Echocardiographic and clinical findings were consistent with cardiogenic shock and were suspicious for ALCAPA. Due to rapid clinical deterioration, MDCT was performed as a fast and accurate diagnostic technique to confirm this diagnosis.

Computed tomography showed the LMCA arising from the posterior and lateral wall of the right pulmonary artery (RPA) and dividing shortly thereafter into the left anterior descending artery (LADA) and circumflex (Cx) artery (Figure 1a). The RCA originated from the right sinus of Valsalva, and it was slightly dilated at 3.2 mm in diameter. The LV and left atrium were also dilated, compressing the right ventricle (RV) (Figure 1b). Based upon these findings, the diagnosis of ALCAPA was established. The patient underwent an operation, and reimplantation of the LMCA into the aorta was performed successfully.

Case 2– *Anomalous LMCA originating from the RCA (Single coronary artery anomaly):* An 11-year-old boy with attacks of palpitation was admitted to the emergency room. Physical examination and chest radiography were normal, but the ECG showed sinus tachycardia. Echocardiography revealed the LMCA arising from the RCA. To confirm this diagnosis, MDCT was performed with a heart rate of 75 beats per minute (bpm) (min: 73, max: 80) without sedation. A CT examination showed the LCA arising from the RCA and taking a course between the aorta and the RV outflow track (Figures 2a and 2b).

Case 3– *A coronary-cameral fistula: An LADA-RV fistula:* A seven-month-old girl was referred to the pediatric cardiology department for evaluation of a cardiac murmur. The patient had no acute respiratory distress or growth retardation. Physical examination revealed a continuous grade 3/6 systolic murmur at the cardiac apex. On echocardiography, a coronary-cameral fistula was suspected. Electrocardiographic-gated coronary CT angiography was requested for further evaluation of the coronary anatomy.

The CT examination was performed without sedation with a heart rate of 126 bpm (min: 124, max: 130). An enlarged tortuous LAD coursed distally and emptied

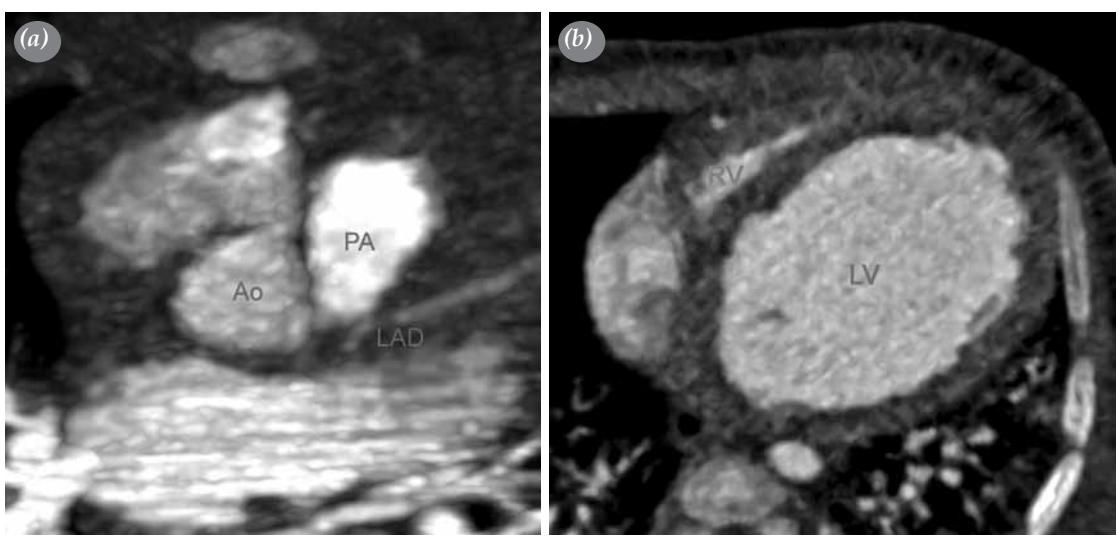

Figure 1. (a) This shows a two-month-old girl with ALCAPA. The three-millimeter axial maximum reconstruction reveals the origin of the left main coronary artery arising from the left inferolateral aspect of the main pulmonary artery and then branching there after into the LAD and circumflex arteries. **(b)** The axial computed tomography shows RV compression from the enlarged LV. PA: Pulmonary artery; Ao: Aorta; LAD: Left anterior descending; RV: right ventricle; LV: Left ventricle.

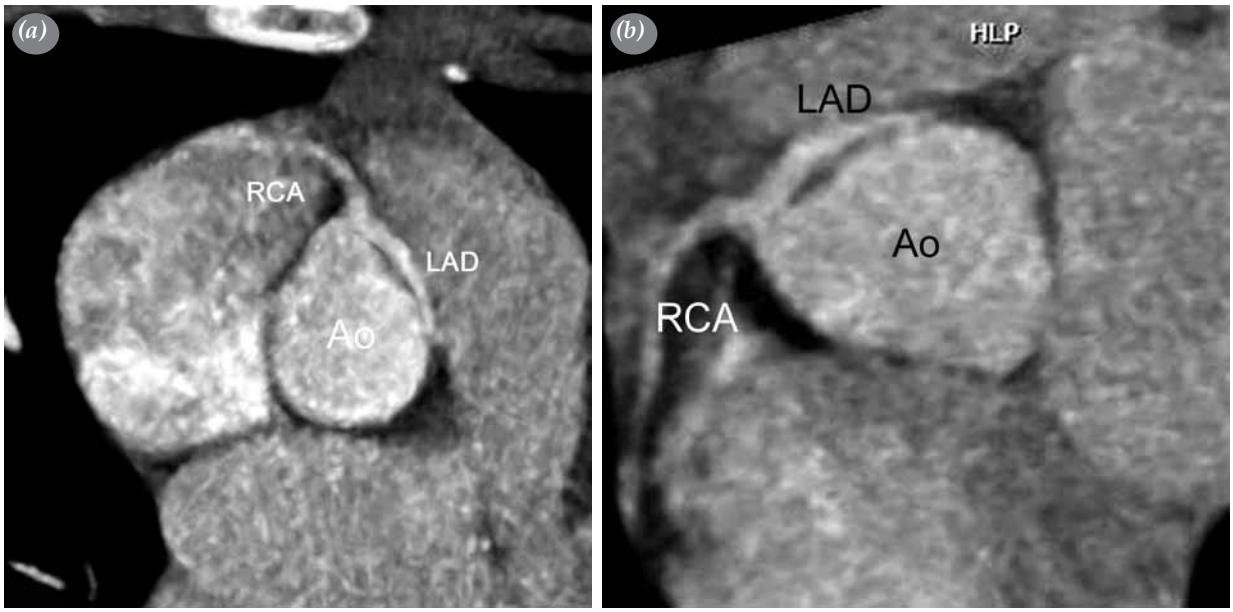

Figure 2. (a) This shows an 11-year-old boy with anomalous origin of the left main coronary artery (LMCA) from the right coronary artery (RCA). The axial computed tomography image shows the LMCA arising from the RCA and taking a course between the aorta and the right ventricle (RV) outflow track. **(b)** A short axis image shows the LMCA arising from the RCA and a slight compression of the LMCA between the aorta and the RV outflow track. RCA: Right coronary artery; LAD: Left anterior descending; Ao: Aorta.

into the RV at the cardiac apex (Figure 3a, 3b). Dilatation of the LCA at 4.3 mm in diameter and that of the LAD at 4 mm in diameter were detected, but the diameter of the RCA was within normal limits with 1.7 mm at the origin.

Case 4– *A coronary-cameral fistula: An RCA-RV fistula in a patient operated on for complex congenital heart disease:* A 10-year-old boy had a history of Fontan operation with the diagnosis of RV hypoplasia as well as tricuspid and pulmonary atresia. A significantly dilated RCA was detected by transthoracic echocardiography. Multidetector CT was performed with the suspicion of a coronary-cameral fistula between the RCA and hypoplastic RV.

Computed tomography showed a dilated and tortuous RCA entering directly into the hypoplastic RV through the apex (Figure 4). The LMCA and Cx were normal in size. The CT examination also demonstrated a hypoplastic RV together with tricuspid and pulmonary atresia with Fontan circulation.

Case 5– *A coronary-cameral fistula: A Cx-coronary sinus fistula in a newborn:* A four-day-old newborn weighing 3.250 grams was referred for a cardiac murmur, dyspnea, and hepatomegaly. A chest roentgenogram demonstrated mild cardiomegaly with normal pulmonary vascularity. Echocardiography demonstrated a Cx-coronary sinus fistula. Also, there was a small mid-muscular ventricular septal defect (VSD) with bidirectional shunt, RV dilatation due to tricuspid regurgitation, and patent foramen ovale. The estimated PA pressure was 65 mmHg.

Multidetector CT was performed to obtain a precise depiction of the coronary artery anatomy, focusing in particular on the anatomic relationship between the Cx artery and the coronary sinus fistula. The examination was completed under sedation with a heart rate of 136 bpm. The CT examination showed that the LMCA orifice was dilated (7 mm). The Cx artery was dilated (5-7 mm) and emptied into the coronary sinus forming the fistula. There was also a severe stenosis at the distal end point of the fistula (Figure 5a). The RCA and LAD were normal in diameter at 2.2 mm and 2 mm, respectively (Figure 5b). There was also a small muscular VSD (Figure 5c).

Case 6– *Anomalous origin of the LMCA from the right sinus of Valsalva:* A 15-year-old boy with recurrent exertional syncope was admitted to our hospital. The physical examination, chest radiography, and ECG findings were normal. Echocardiography revealed a suspicion of an anomalous origin of the LMCA from the right coronary sinus. In the exercise testing, the patient had developed a syncope attack with hypotension, bradycardia, and ischemic findings in the ECG.

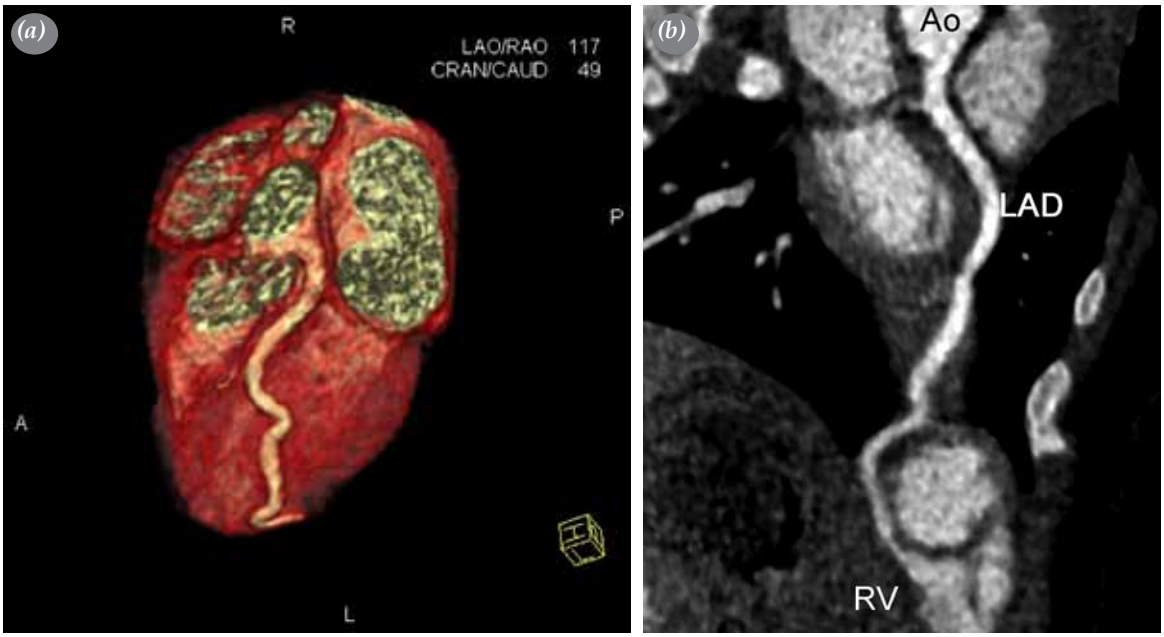

Figure 3. (a) This shows a seven-month-old infant with an left anterior descending (LAD)-right ventricle (RV) fistula. The 3D-volume rendered technique (VRT) image shows the enlarged tortuous LAD coursing distally and emptying into the RV at the cardiac apex. **(b)** The curved, multiplanar, reformatted image shows the origin and end point of the LAD clearly. LAD: Left anterior descending; Ao: Aorta.

To confirm this diagnosis, MDCT was performed with a heart rate of 75 bpm (min: 73, max: 80). Computed tomography showed the LMCA arising from the right sinus of Valsalva with an interarterial course (Figure 6a). The RCA had a separate ostium directly from the right sinus of Valsalva. Reconstruction of the LMCA origin with an autologous pericardial patch was performed successfully. A follow-up postoperative CT examination was performed (Figure 6b).

RESULTS

Three anomalous origin (one ALCAPA, two anomalous left main coronary artery originated from right sinus of Valsalva) and three fistulas (two coronary-cameral fistula, one left main coronary artery -coronary sinus fistula) were clearly identified with dual-source MDCT angiography.

DISCUSSION

The primary congenital anomalies of the coronary arteries have an incidence of 1-2% in the general population.[9] Although many of these anomalies are benign, a small number of them carry the risk for ischemia and sudden death.[10]

Until recently, conventional coronary angiography was the diagnostic method of choice for detecting coronary artery anomalies, but the precise course and anatomic relationship of the anomalous vessel could be difficult to delineate because of the two-dimensional (2D) imaging capability of the conventional coronary

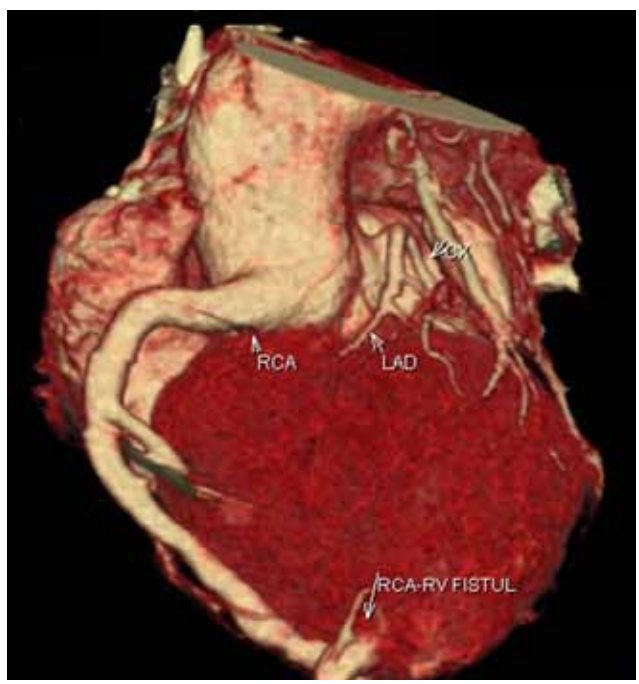

Figure 4. A 10-year-old-boy having a history of Fontan operation had a coronary-cameral fistula. The 3D-VRT image shows the dilated and tortuous right coronary artery entering directly into the cavity of the right ventricle through the apex.

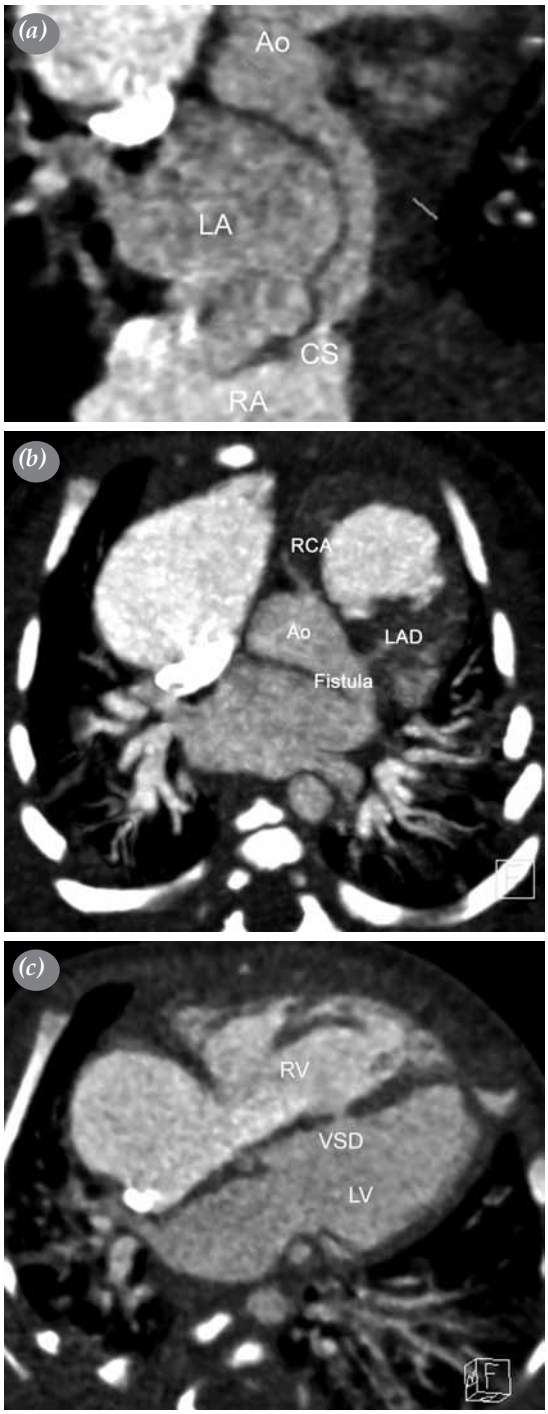

Figure 5. (a) This shows a four-day-old infant with a circumflex (Cx)-coronary sinus fistula. Curved multiplanar reformatted (MRP) images show the left main coronary artery and Cx arteries were dilated, and they emptied into the coronary sinus with a severe stenosis at the distal part of the fistula. **(b)** In the axial maximum intensity projection (MIP) image, the RCA and LAD were normal in size. **(c)** In the four chamber MIP image, there is a small VSD. Ao: Aorta; LA: Left atrium; CS: Coronary sinus; RA: Right atrium; RCA: Right coronary artery; LAD: Left anterior descending; RV: Right ventricle; VSD: Ventricular septal defect; LV: Left ventricle.

angiography. Although MDCT is still a developing technology for imaging of the coronary arteries, it is superior to conventional angiography in the diagnosis of coronary anomalies and fistulas given the multiplanar and three-dimensional (3D) imaging capabilities that allow for the precise definition of spatial relations of the anomalies and fistulas in children.[11,12]

Present in one out of every 300,000 live births, ALCAPA is a rare congenital anomaly usually seen as an isolated lesion. There are two types: the infant type and the adult type, and it represents one of the most common causes of myocardial ischemia and infarction in childhood. Soon after birth, blood flow in the LCA is reversed emptying into the PA, a condition known as the "myocardial steal" syndrome. Due to little or no coronary collateral development in infants, this condition leads to the early onset of severe myocardial ischemia, LV dysfunction, and dilatation if left untreated, with a mortality rate of up to 90% within the first year of life. The most important differential diagnosis in this age group is with dilated cardiomyopathy, which also commonly presents with moderate to severe congestive heart failure, massive cardiomegaly, and ischemic signs on the ECG.[13,14]

Electrcardiographic-gated MDCT angiography plays an important role as an alternative modality in the assessment of ALCAPA, especially in patients with cardiogenic shock or severe heart failure, as was the case in one of our patients. A characteristic MDCT angiography finding of this condition is an LCA typically arising from the left inferolateral aspect of the main PA just beyond the pulmonary valve. The LMCA is markedly dilated and tortuous in adult patients but appears normal in size in infants. In our patient, the LMCA and RCA were not dilated.

Another type of anomalous origin of coronary arteries is the LCA arising from the right sinus of Valsalva as a separate vessel or as a branch of a single coronary artery. It has an incidence of 0.09-0.11% in conventional angiography. An interarterial course may be seen in up to 75% of these patients, who run a high risk for sudden death due to the acute angle of the ostium, the stretch of the intramural segment, and the compression between the right and left coronary cusps. This lethal coronary arterial malformation is rarely identified and is usually only identified during an autopsy. Multidetector CT plays an important role in the diagnosis of this pathology.[15-18] In case 2 and case 6, anomalous origin of the LCA with its 3D relationships was clearly demonstrated with MDCT, and there was no need for catheter angiography.

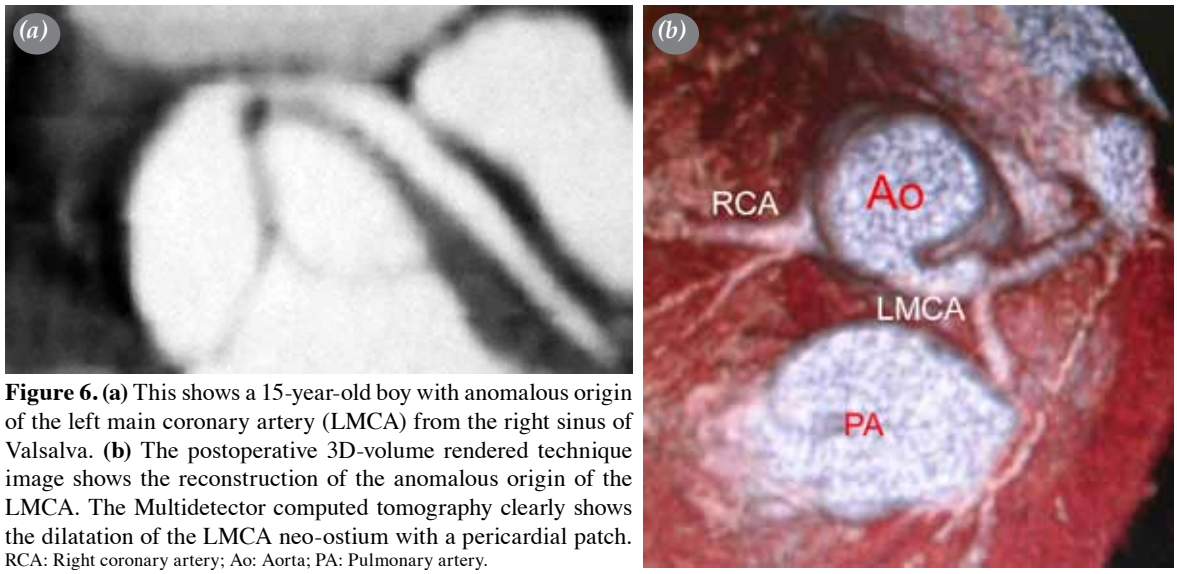

Figure 6. (a) This shows a 15-year-old boy with anomalous origin of the left main coronary artery (LMCA) from the right sinus of Valsalva. **(b)** The postoperative 3D-volume rendered technique image shows the reconstruction of the anomalous origin of the LMCA. The Multidetector computed tomography clearly shows the dilatation of the LMCA neo-ostium with a pericardial patch. RCA: Right coronary artery; Ao: Aorta; PA: Pulmonary artery.

Multidetector CT also plays an important role in the postoperative evaluation of the LMCA. In case 6, postoperative CT clearly demonstrated the reconstructed LMCA ostium and its relationship with the aortic root and PA.

A coronary artery fistula is another type of congenital coronary anomaly with an incidence of 0.2-0.4%, and it occurs when communication is present between a cardiac chamber and a coronary artery. The involved coronary artery is dilated due to the increased blood flow and is often tortuous to an extent determined by shunt volume. In 90% of cases, it poses a shunt from the high-pressure coronary artery to a lower-pressure cardiac chamber or vein. Myocardial perfusion may be decreased in the involved myocardium supplied by the abnormally connecting coronary artery.[11,12,19] Dilatation of the involved coronary artery makes it easier to demonstrate via MDCT, even in infants with high heart rates. Computed tomography angiography was completed successfully without a beta-blocker and sedation in a seven-month-old patient with a coronary-cameral fistula. In a patient with Fontan operation due to tricuspid atresia, MDCT was depicted not only in the RCA-hypoplastic RV fistula, but it was also demonstrated in the Fontan anastomosis and cardiac chambers, which would not have been possible with conventional angiography and echocardiography. Because the long-term complications of cyclophosphamide (CPA), adriamycin (ADR) and 5-fluorouracil (5-FU) (CAF) treatment may include coronary thrombosis, myocardial infarction, and cardiomyopathy, proper preoperative evaluation with MDCT is crucial for planning an optimal therapeutic strategy.[12]

Concerns about the radiation dosage and higher heart rates cause limitations with regard to the use of CT for coronary artery evaluation in children and infants. Dual source MDCT, with a high temporal resolution independent of the heart rate, is also superior to the older generation CT scanners in coronary artery imaging. A dual source CT has two X-ray tubes as well as two corresponding detectors that are arranged in a perpendicular configuration. This configuration provides the temporal resolution to fall as low as 83 ms independent of the patient's heart rate. Improved temporal resolution decreases the motion artifact, thus improving, even at higher heart rates, the accuracy of the coronary artery visualization without the need of beta-blockers.[1,20-23] In all our patients, in spite of the high heart rate (136 bpm), optimal diagnostic images were obtained without the requirement of beta-blockers.

The automatic adaptation of pitch is another important advantage of dual source scanners. As the heart rate increases, the table speed increases, which significantly decreases the radiation dose to the patient.[1]

In conclusion, coronary MDCT angiography is an accurate and noninvasive tool for delineating coronary artery anatomy before surgical intervention in children with or without other associated congenital cardiac anomalies. Dual source MDCT is a significant alternative to conventional coronary angiography and can be used routinely in combination with clinical, echocardiographic analysis for the preoperative assessment of patients. Regarding radiation concerns,

examination performed with as low as reasonably achievable (ALARA) principles decreases the amount of radiation significantly without affecting the diagnostic image quality.

Declaration of conflicting interests

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