ORIGINAL ARTICLE

Prevalence and characteristics of coronary artery anomalies in children with congenital heart disease diagnosed with coronary angiography

Doğuştan kalp hastalığı olan çocuklarda anjiyografiyle belirlenmiş koroner arter anomalilerinin prevalansı ve karakteristiği

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ABSTRACT

Objective: Aim of the present study was to determine the prevalence of coronary artery anomalies in children with congenital heart disease.

Methods: Data of 1138 consecutive patients who were referred for cardiac catheterization and angiography for assessment of coronary anomaly between January 2005 and December 2009 were retrospectively analyzed. Total of 515 patients whose coronary arteries could be examined through left ventricle and aortic root injection were included in the study.

Results: Of 515 angiograms with visible coronaries, 42 patients (20 males, 22 females; mean age: 5.3±2.0 years) were found to have final diagnosis of coronary anomaly. Prevalence of coronary artery anomalies was 8.16% in this study. It was determined that 38 (90.4%) were anomalies of origination, 2 (4.8%) were anomalies of intrinsic coronary arterial anatomy, and 2 (4.8%) were anomalies of coronary termination. Most common coronary artery abnormality was anomalous origin of the right coronary artery from the left aortic sinus (16 patients; 38.1%), and the most common congenital heart disease was tetralogy of Fallot (18 patients; 42.9%).

Conclusion: Recognizing variability of coronary artery anomalies is critical when considering surgical or interventional therapies in children with congenital heart disease.

ÖZET

Amaç: Bu çalışmanın amacı, doğuştan kalp hastalığı olan çocuklarda koroner arter anomalilerinin sıklığını belirlemektir.

Yöntemler: Ocak 2005 ve Aralık 2009 tarihleri arasında kardiyak kateterizasyon ve anjiyografi için başvuran toplam 1138 hasta koroner anomali açısından geriye dönük olarak değerlendirildi. Çalışmaya sol ventrikül ve aort kökü enjeksiyonlarıyla koroner arter görüntülemesi yapılabilen toplam 515 hasta alındı.

Bulgular: Koroner görüntülemesi yapılan 515 hastanın 42'sinde (20 erkek, 22 kız; ortalama yaş 5.3±2.0 yıl) koroner arter anomalisi tespit edildi. Çalışmada koroner arter anomali prevalansı %8.16 olarak bulundu. Koroner arter anomalilerinin 38'i (%90.4) orijin, ikisi (%4.8) intrinsik ve ikisinin (%4.8) sonlanım anomalisi olduğu belirlendi. Koroner arter anomalileri arasında en sık sol aortik sinüsten kaynaklanan sağ koroner arterin orijin anomalisi (16 hasta, %38.1) olduğu, doğuştan kalp hastalıkları arasında ise en sık görülenin Fallot tetralojisi (18 hasta, %42.9) olduğu tespit edildi.

Sonuç: Doğuştan kalp hastalığı olan çocuklarda cerrahi veya girişimsel tedavi düşünülürken, koroner arter anomalilerinin çeşitliliğinin fark edilmesi kritik önemdedir.

Congenital coronary artery anomalies (CAA) are of major significance in clinical cardiology and cardiac surgery due to association with life-threatening symptoms, including arrhythmias, syncope, myocardial infarction, and sudden death.[1] Identification of CAA can be documented by autopsy, echocardiography, diagnostic coronary angiography, multislice coronary computed tomography (CT) angiography,

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or cardiac magnetic resonance imaging. Currently, the most commonly used classification of CAA is based purely on anatomical considerations, recognizing 3 categories: (1) anomalies of origin and course, (2) anomalies of intrinsic coronary artery anatomy, and (3) anomalies of termination.[2,3] Coronary artery anomalies vary with respect to number, location, orientation of the ostia, and origin of the coronary arteries. Some anomalies are merely anatomical variants without clinical relevance, while others may be life-threatening. Undiagnosed anomalous coronary arteries are a well-recognized cause of sudden cardiac death in children.[4] CAA comprises 20% of cases of sudden cardiac death in young athletes.[5]

CAA may be associated with complex congenital heart disease, but may also occur as isolated anomaly. In patients undergoing coronary angioplasty or cardiac surgery, angiographic recognition of coronary anomalies is important for proper management of these patients. Overall prevalence of isolated coronary artery anomalies in various studies ranges from 0.3% in a study by Akpinar et al.,[6] using conventional catheter angiography, to an incidence of approximately 18.5% in a study conducted by Cademartiri et al.,[7] based on 64-slice coronary CT angiography. In 1 recent study, incidence of CAA was found to be 0.9% in a pediatric population.[8] Among those with symptomatic congenital heart disease, coronary artery anomalies are much more common and occur in approximately 11% to 14% of cases.[9,10] Routine preoperative demonstration of coronary artery anatomy in tetralogy of Fallot patients can be achieved satisfactorily and conveniently with coronary angiography.[11] Reported incidence of coronary anomalies in cases of tetralogy of Fallot has varied widely, depending on method of detection.[11–13]

The purpose of this study was to retrospectively determine prevalence of CAA in children with congenital heart disease who presented with different symptoms and underwent coronary angiography for assessment of coronary artery disease.

METHODS

Between January 2005 and December 2009, total of 1138 consecutive patients were referred to tertiary medical center for diagnostic, emergency, or elective coronary angiography due to suspicion of congenital heart disease. Datasets of these patients were retrospectively reviewed in search of coronary anomalies of origination and further vessel course. Of 1138 coronary cineangiograms, 515 patients whose coronary anatomy could be completely defined were included in this study. Evaluation of these 515 arteriographies, which included both intravenous and aortic root angiography, detected coronary anomalies and diagnoses were confirmed.

Abbreviations:

CAA	*Congenital coronary artery anomalies*
CT	*Computed tomography*
LAD	*Left anterior descending*
LCA	*Left coronary artery*
LCX	*Left circumflex*
LMCA	*Left main coronary artery*
RCA	*Right coronary artery*

The following were accepted as coronary variants and not included in the final analysis: separate conal branch from the right coronary artery, "high take-off" right coronary artery or left main stem from the aorta, separate left anterior descending (LAD) artery, left circumflex (LCX) artery from the aorta with absent left main stem. Patients with truncus arteriosus or transposition of the great arteries were also excluded from the study. In addition, coronary abnormalities not hereditary in origin, such as coronary aneurysm, were also excluded. Final analysis included 42 patients with definite diagnosis of coronary artery anomaly.

RESULTS

Coronary angiograms were reviewed for presence of coronary anomaly in at least 1 major epicardial coronary artery. All indications for arteriography were related to evaluation of the heart during diagnostic, emergency, or elective coronary angiography. Total of 42 patients diagnosed with coronary anomaly were found among 515 angiography reports with visible coronaries. Angiography records of these patients were retrieved from electronic medical database and reviewed for coronary anomalies. All of the patients had congenital heart disease. Mean age was 5.3±2.0 years (range: 1–9 years). There were 22 (52.4%) females and 20 (47.6%) males. Number of patients with origination, course, and termination anomalies were determined to be 38 (90.4%), 2 (4.8%), and 2 (4.8%), respectively (Table 1).

Anomaly was categorized into 10 major groups according to origin, course, or distribution of the involved major epicardial coronary artery for all patients in the study group. In addition to specific anomaly, patients were further divided into 2 major groups according to

Table 1. Coronary artery anomaly characteristics of the patients

	Number of patients	
	n	%
Coronary artery anomalies		
Origin	38	90.4
Course	2	4.8
Termination	2	4.8
Total	42	100

Data are mean±SD.

the Angelini classification.[2] Details of these coronary artery anomalies are summarized in Table 2. Right coronary artery (RCA) and left main coronary artery (LMCA) arising from left aortic sinus were found to be the most common anomalous vessels, involved in 16 (38.1%) patients as origination anomaly. Anomalous ostium near sinotubular junction was the second most common anomaly, with 7 patients (16.6%). Single coronary artery arising from the left sinus and abnormal number of ostia was the third most common category of anomaly, with 4 patients (9.5%). Among the 515 patients, 42 were diagnosed with CAA; overall incidence of CAA in study population was 8.16% (Table 2). In the present study, 5 (11.9%) single coronary arteries were found. One (2.4%) originated from right aortic sinus, and the other 4 (9.5%) from the left aortic sinus, with none coursing between the great arteries. Two patients (4.8%) with single coronary artery had tetralogy of Fallot, 2 (4.8%) had aortic stenosis, and 1 (2.4%) had double outlet right ventricle.

In this study, no coronary artery originating from the pulmonary artery, coronary artery aneurysm, stenosis, or bridging was found. Most common heart disease was tetralogy of Fallot among 42 patients with

Table 2. Coronary artery anomalies, number of patients, and angiographic incidence

Coronary artery anomalies	n	Anomaly incidence (n=42) (%)	Angiographic incidence (n=515) (%)
1. Origin anomalies			
A. Anomalous pulmonary origin			
B. Anomalous origin from aorta			
1. LMCA and RCA from right sinus	2	4.8	0.38
2. LMCA and RCA from left sinus	16	38.1	3.10
3. LMCA and RCA from posterior sinus	3	7.1	0.58
4. RCA and Cx from right, LAD from left sinus	1	2.4	0.19
C. Single coronary artery arising from aorta			
1. From right sinus	1	2.4	0.19
2. From left sinus	4	9.5	0.77
D. Ostium anomalies			
1. Abnormal number of ostia	4	9.5	0.77
2. Abnormal ostium near sinotubuler junction	7	16.6	1.35
2. Course anomalies			
A. Coronary artery aneurysm			
B. Coronary stenosis			
C. Hypoplasia of the coronary artery			
D. Coronary artery bridging	2	4.8	0.38
3. Termination anomaly			
A. Arteriovenous fistula	2	4.8	0.38
Total	42	100.0	8.16

LMCA: Left main coronary artery; RCA: Right coronary artery; Cx: Circumflex; LAD: Left anterior descending.

Table 3. Distribution of congenital heart disease among 42 patients with coronary anomalies

Congenital heart diseases	n	%
Tetralogy of Fallot	18	42.9
Double outlet right ventricle	8	19.0
Aortic stenosis	5	11.9
Ventricular septal defect	4	9.5
Patent ductus arteriosus	3	7.1
Atrial septal defect	2	4.8
Coronary artery fistula without intracardiac pathology	2	4.8

CAA (Table 3). Of 121 patients diagnosed with tetralogy of Fallot among initial 515 coronary angiography reports, coronary anomaly incidence was determined to be 14.8%, with tetralogy of Fallot observed in 18 of 121 patients. Abnormal conal branch was accepted to be variant of normal in 6 (33.3%) patients with tetralogy of Fallot. Patient with aneurysm of sinus of Valsalva, and 2 patients with pericardial collateral artery originating from abdominal aorta were excluded because these aberrations were outside of any classification. In addition, patient with dual LAD artery was excluded because this anomaly was not part of the classification used.[2]

DISCUSSION

The exact prevalence of congenital coronary anomalies in the general population remains uncertain. Previous studies have suggested that these malformations occur in 0.3% to 18.5% of large group of patients whose coronary angiography has been studied.[6,7] Proposed prevalence varies, depending on diagnostic criteria, method used to diagnose coronary anomaly (e.g., autopsy, transthoracic echocardiography, coronary angiography, or CT angiogram), and the population studied, which is typically a targeted population, such as athletes[5] or children.[8] We have described our experience with congenital CAA diagnosed by coronary angiography in a series of pediatric patients. Incidence of 8.16% was determined in the present study. Chen et al.[10] used neonates/infants/children/adolescents in their CT study, and reported 13.6% incidence of CAA. Several studies using both children and adults have reported incidence of CAA with congenital heart disease of 0.16%[14] and 11%.[9]

Anomalies involving abnormal origin of the coronary arteries are clinically important and are commonly associated with some form of congenital heart disease. Presence of coronary artery anomalies in the absence of any other congenital heart defect is considered rare (in 0.5% of pediatric autopsies).[15] Anomalous origin of left coronary artery (LCA) from the pulmonary artery is a rare anomaly that occurs in approximately 1 per 300,000 live births and represents 0.5% of all congenital heart defects.[16] Congenital hypoplastic, stenotic, or atretic left main coronary artery (LMCA) is rare anomaly, and has been found to be related to myocardial infarction, but not sudden death.[17] There is wide variety in reported prevalence rate of CAA. Incidence of major epicardial coronary artery anomaly was found to be 0.3% in a retrospective study.[6] Overall incidence of primary congenital coronary anomalies reported as 0.8% among patients undergoing diagnostic coronary arteriography.[18] In the literature, the largest study on coronary anomalies was reported by Yamanaka and Hobbs.[19] They reported 1.3% incidence of CAA. De Jonge et al.[20] described prevalence of 7% of CAA, including coronary fistulas, in their patient population. Tongut et al.[21] reported that prevalence of coronary anomalies was 9.37%, including all types of coronary anomalies with specified classification. Yu et al.[22] demonstrated that incidence of major CAA was 15.11% in patients ranging in age from 1 month to 76 years. Few studies have investigated CAA in the pediatric population, and incidence of CAA is reported to be 0.7% to 1.4%.[23–25] Angelini et al.[25] reported on screening magnetic resonance imaging study of 3165 adolescents (aged 11–18 years) and found 22 cases (0.7%) of CAA. Present study found higher incidence (8.16%) of CAA in children with congenital heart disease.

According to the current literature, majority of coronary anomalies are associated with origination or distribution (course, structure) of a coronary artery, whereas anomalies of termination (fistula, etc.) are relatively less frequent.[2] It has been reported that aberrant arteries are 90% origin and course, and 10% termination-related.[18,26] Consistent with the literature, majority of coronary anomalies observed in the present study were associated with origin of the coronary arteries. We found that 90.4% of anomalies were related to origination, and 4.8% were termination anomalies.

Anomalous coronary artery from the opposite sinus includes RCA arising from the left sinus and LCA arising from the right sinus. Most common finding is LCX arising from the right sinus.[27] This is followed by RCA arising from left sinus, and lastly, LMCA arising from the right sinus.[27] However, RCA arising from left sinus was the most common anomalous artery, with 16 patients, representing an incidence of 3.1%, LMCA arising from the right sinus was detected in 2 patients in our study, an incidence of 0.38%. These results were similar to ratio found in angiographic series conducted by Angelini et al.[28] Yamanaka and Hobbs[19] also noted that anomalous origin with RCA arising from the left coronary sinus (0.17%) is far more common than LCA from the RCA sinus (0.047%). LCX arising from right sinus was detected in only 1 patient, an incidence of 0.19%. LMCA and RCA arising from posterior aortic sinus is another rarely seen, wrong-sinus-origin anomaly. Yamanaka and Hobbs[19] found this ratio to be 0.3% of all anomalies. In our study, we found this ratio to be 7.1%, a total of 3 patients.

Ostium abnormalities are abnormal number or position of the ostia. Yamanaka and Hobbs[19] reported ostium anomalies as most frequent and examined it as separated ostia of LAD and LCX arteries arising from left sinus of Valsalva. In our study, ostium anomalies were the second most frequent anomaly, with ratio of 26.1% in 11 patients. Coronary hypoplasia is a rarely seen and not well-defined anomaly. In our study, there were 2 cases, representing ratio of 4% of all anomalies. Coronary artery fistula is an anomaly of termination.[29] Fistula can cause pulmonary hypertension, congestive heart disease, and bacterial endocarditis, and size of the fistula and number of shunts determine the hemodynamics. Coronary artery fistula is rare form of coronary artery anomaly, and prevalence was reported as 0.01% to 0.2% in adults.[30–32] In our study, there were 2 children with fistula, a ratio of 4.7%.

Coronary anomalies appear more often in certain congenital heart diseases. Coronary anomaly incidence has been reported as 2% to 14% in cases of tetralogy of Fallot.[11–13,33] This wide range difference is result of dilated conal branch coursing to right ventricular outflow tract. It is usually misevaluated as coronary anomaly. It has been determined that coronary artery anomaly incidence is 12.7% overall, using combination of angiographic and surgical methods in children with tetralogy of Fallot.[34] In our study, coronary anomaly incidence was 14.8%, with 18 of 121 patients diagnosed with tetralogy of Fallot.

In conclusion, results of the present study demonstrated 8.16% incidence of coronary anomaly in children with congenital heart disease. Cardiologists and surgeons should be familiar with CAA for proper management of patients undergoing cardiac surgery or coronary angioplasty.

Conflict-of-interest: None declared.

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