

Assessment of Cardiovascular Surgery Requirements in Children and Adolescents Admitted With Chest Pain To A Pediatric Emergency Clinic

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ABSTRACT

Introduction: Chest pain in children and adolescents is an important symptom in the pediatric emergency clinic. The aim of this study is to assess the characteristics of chest pain and cardiovascular surgery in the pediatric population.

Materials and methods: There were 352 children who presented with chest pain to the pediatric emergency department between December 2007 and February 2017. These children were included in this study, which is a retrospective observational study.

Results: Among the 352 patients, six patients (1.7%) underwent cardiovascular surgery. Forty-eight patients (13.6%) were diagnosed with cardiac disease, and the most common cardiac causes were myocarditis and pericarditis.

Conclusion: Only six patients (1.7%) who were admitted to the emergency department with chest pain needed cardiovascular surgery. The chest pain in children required less surgery, and mortality and morbidity were lower compared with adult cardiac chest pain.

INTRODUCTION

Chest pain is common in children and adolescents and ranks third after headache and abdominal pain among the reasons for consulting a doctor [Massin 2004]. Complaints of chest pain were in 0.3-0.6% of the patients admitted to the pediatric emergency clinic [Evangelista 2000]. The association of chest pain with angina pectoris and myocardial

infarction in adult patients is a well-known situation, however, this is not common in children and adolescents. On the other hand, chest pain in children may be a sign of serious heart conditions that should be differentiated [Evangelista 2000; Sert 2013]. The aim of this study is to investigate the characteristics of the chest pain, surgical requirement, and techniques in the pediatric population.

MATERIALS AND METHODS

This retrospective study was conducted with the approval of the institutional review board and ethics committee of our university hospital. Ten years of data were investigated, and 352 cases were included in this study. These patients were admitted with chest pain to the pediatric emergency outpatient clinic between December 2007 and February 2017. The patients ≤ 18 years old included in this study; patients who underwent cardiac surgery for previously diagnosed congenital heart disease were excluded from the study. Medical records were available in the hospital archive.

The characteristics of the chest pain, symptoms accompanying pain (palpitations, fainting, dizziness/darkening of the eyes, cough, shortness of breath, abdominal pain, fever, weakness, sweating), and physical examination findings were recorded from hospital records.

Records of patients with suspected cardiac pain and undergoing further evaluation (echocardiography, telecardiography, cardiac magnetic resonance imaging, coronary CT angiography, coronary angiography, and 24-hour rhythm Holter monitoring) were examined. In our hospital, we have the opportunity to perform echocardiography for 24 hours. Echocardiography was performed on almost all patients in this study group except the patients with overt musculoskeletal pain. Furthermore, cardiac magnetic resonance imaging was performed in 11 patients, coronary computed tomography (CT) in nine patients, thorax CT in eight patients, myocardial perfusion imaging in one patient, coronary angiography in eight patients, 24-hour Holter monitoring in 55 patients, event recorder in one patient, and exercise tests in 38 patients. The patients who underwent

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surgical treatment and the type of the surgical procedures are discussed in detail.

RESULTS

The mean age of patients with cardiac cause was 12.4 ± 3.1 years and after exclusion of an apparent cause, 51.1% of the patients ($N = 180$) were diagnosed as idiopathic. Forty-eight (13.6%) patients were diagnosed with cardiac disease, 49 (13.9%) patients were diagnosed with musculoskeletal system disease, 33 (9.3%) patients were diagnosed with psychogenic disease, 22 (6.2%) patients were diagnosed with gastrointestinal disease, and 18 (5.1%) patients were diagnosed with respiratory disease as a cause of chest pain. (Figure 1) Among the cardiac diseases, 23 patients (47.9% of total cardiac causes) were diagnosed with pericarditis and myocarditis. Chest pain was exacerbated by lying down in 91% of these patients, and 83% of them had fever (> 38 C, tympanic measurement). Pericardiocentesis was successfully performed in seven patients. Mitral valve prolapse (18.7%) and myocardial bridging (10.4%) were the third and fourth cardiac cause of the chest pain, respectively, and they followed up medically. The most affected artery in patients with myocardial bridge was the left anterior descending artery, and they were diagnosed with computed tomography (Figure 2A, B). (Figure 2) One patient was diagnosed with hypertrophic cardiomyopathy and was recommended implantable cardioverter defibrillator implantation, due to history of sudden cardiac death in her family. Two patients, who were previously diagnosed with Duchenne muscular dystrophy (DMD), presented with chest pain. Coronary angiography showed normal coronary arteries in both patients, and they were discharged without any problem. Acute myocardial injury due to DMD was considered. One patient was admitted with supraventricular tachycardia and later diagnosed with wolf parkinson white syndrome. It was consulted with the electrophysiology department for further evaluation. (Table 1)

Chest pain was characterized with sharp-like stabbing in most of the patients ($N = 211$, 59.9%) and pressure-like chest pain was found in 96 (27.2%) patients. The location of the pain was at the left side of the sternum 71.8% ($N = 253$) of the patients and at the middle of the chest in 12.2% ($N = 43$) patients. (Table 2)

Palpitation was the most relevant symptom (19%) associated with chest pain among the 352 patients. The other most common symptoms associated with chest pain were dyspnea (12.5%) and dizziness (7.6%). Only one patient admitted with cardiac arrest was successfully resuscitated. (Table 3)

Among the 352 patients, 61% of them ($N = 215$) had resting chest pain. The chest pain increased with exercise in 13% ($N = 46$) and increased with breathing in 14.2% ($N = 50$) of them. Most of the patients ($N = 21$, 43.7%) with cardiac disease also had resting chest pain and 25% ($N = 12$) of them had increased chest pain with exercise. (Table 4)

Cardiovascular surgery was performed in six patients (1.7%) of total 352 patients who presented with chest pain.

The patients, who underwent cardiovascular surgery, were diagnosed with ALCAPA syndrome, Kawasaki disease, Homozygous FH, Loey-Dietz Syndrome, rheumatic mitral insufficiency, and RCA arising from the left coronary ostium. (Table 5)

Troponin level was available in 318 patients and 25 of these patients were diagnosed with cardiac disease (11 myocarditis, three myocardial bridging, two acute coronary syndrome, two acute myocardial injury due to Duchenne muscular dystrophy, one anomalous left coronary artery from pulmonary artery (ALCAPA) syndrome, one coronary artery anomaly, and one dilated cardiomyopathy).

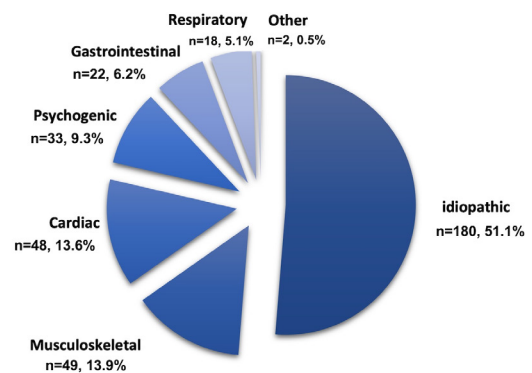


Figure 1. The percentage of diseases that caused chest pain in children who were admitted to the emergency department.

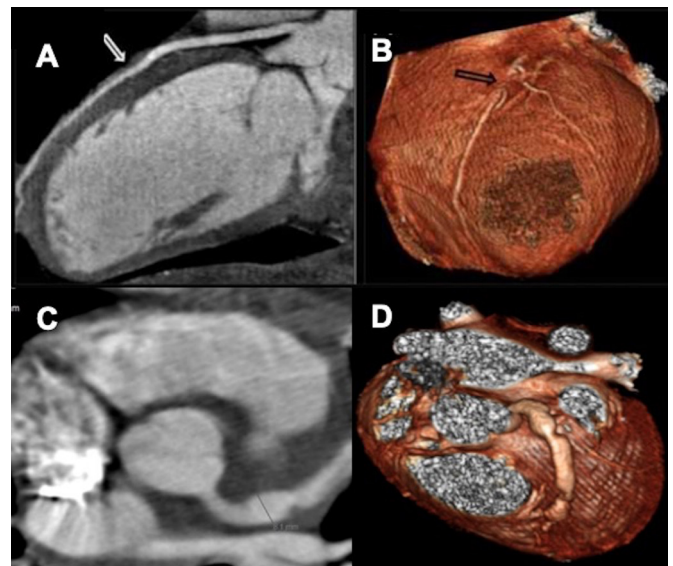


Figure 2. Computed tomography angiography (with three-dimensional volume rendering) showing myocardial bridging in proximal left anterior descending artery (A, B); computed tomography angiography (with three-dimensional volume rendering) revealing left anterior descending artery aneurysm in Kawasaki disease (C, D).

DISCUSSION

In this study, we found that 13.6% of the pediatric patients had cardiac disease who were admitted to the pediatric emergency department with chest pain. Six patients (1.7% patients with chest pain) underwent successful cardiovascular surgery after careful assessment.

Pediatric chest pain is a concern for the parents, due to mortality and morbidity information in the adult patient group, however, chest pain is less likely to be of cardiac origin than adults. Most chest pain etiology in the childhood period are benign in character and include the musculoskeletal system and psychogenic origin [Aygün 2020]. The most common cardiac etiology was pericarditis and myocarditis, which successfully were treated with medical therapy. Seven patients required pericardiocentesis during in-hospital follow up. Moreover, as seen in our study, the patients who require cardiac surgery successfully can be treated with a meticulous

diagnostic algorithm. No cardiac mortality occurred in this study with subjects who presented with chest pain, despite additional surgical interventions in the following periods. In the literature, there was not much data about this ratio and approximately two children in 100 patients may require cardiovascular surgery.

Coronary artery disease due to atherosclerosis in childhood is extremely rare. Familial hypercholesterolemia occurs due to gene mutations related to low density lipoprotein receptors, which results in an increase in low density lipoprotein levels [Berenson 1998]. In cases of homozygous familial hypercholesterolemia, the aortic valve, ascending aorta, and the coronary arteries can be affected. A 15-year-old female patient presented with a complaint of chest pain accompanied by dyspnea that increased with effort and spread to the back, jaw, and left arm for a period of two days. The patient previously was diagnosed with homozygous familial hypercholesterolemia; however, she did not have regular hospital

Table 1. Cardiac causes and clinical characteristics of the chest pain in the pediatric population admitted to our emergency department

Diagnosis	Number of patients (N = 48)	Time of diagnosis	Age, mean (min-max)	Clinical findings	Diagnosis modality	Management
Pericarditis- myocarditis	23 (47.9%)	Newly	14.3 (8-18 years)	Weakness, fatigue, fever increased CP with lying	Lab, ECG, TTE	Medical, seven patients underwent pericardiocentesis
Mitral valve prolapse	9 (18.7%)	Newly	13 (7-17 years)	Palpitation, murmur	TTE	Medical
Myocardial bridging	5 (10.4%)	Newly	13.8 (7-18 years)	Severe CP	CT	Medical
Acute coronary syndrome due to FH	1 (2%)	Previously diagnosed for FH	15 years	CP, exertional dyspnea	Lab, ECG, CT, CA	Surgery, lipid apheresis
Acute coronary syndrome due to KD	1 (2%)	Diagnosed two years ago for KD	9 years	CP, ECG	Lab, CT, MPS	Surgery
Myocardial injury due to DMD	2 (4.1%)	Previously diagnosed for DMD	14 years, 15 years	CP	ECG, CA	Medical
ALCAPA	1 (2%)	Newly	16 years	Cardiac arrest	ECG, TTE, CA	Surgery
Aortic root aneurysm due to Loeys-Dietz Syndrome	1 (2%)	Newly	11 years	CP, palpitation, murmur	TTE	Surgery
Single coronary artery	1 (2%)	Newly	17 years	CP	CT	Surgery
Hypertrophic cardiomyopathy	1 (2%)	Newly	-	CP, exercise intolerance, murmur	ECG, TTE	Medical, ICD recommended
Dilated cardiomyopathy	1 (2%)	Newly	14 years	Dyspnea, CP, weakness	Lab, ECG, TTE	Medical
Rheumatic mitral insufficiency	1 (2%)	Previous acute rheumatic fever	16 years	CP, exercise intolerance	ECG, TTE	Surgery
Wolf Parkinson White	1 (2%)	Newly	12 years	CP, palpitation	ECG	Electrophysiologic study

*FH, familial hypercholesterolemia; KD, Kawasaki disease; DMD, Duchenne muscular dystrophy; ALCAPA, anomalous left coronary artery from pulmonary artery; CP, chest pain; Lab, laboratory; ECG, electrocardiography; TTE, transthoracic echocardiography; CT, computed tomography; CA, coronary angiography; ICD, implantable cardioverter defibrillator

Table 2. Characteristics of the chest pain of the study subjects

	N = 352
Chest pain quality	
Sharp, pinprick	211 (59.9%)
Pressure	96 (27.2 %)
Squeezing	25 (7.1 %)
Burning	11 (3.1 %)
Aching	9 (2.5%)
Chest pain localization	
Left anterior chest wall	253 (71.8%)
Right anterior chest wall	5 (1.4%)
Middle of the chest	43 (12.2%)
Left and middle of the chest	32 (9.1%)
Epigastric	19 (5.4%)

Table 3. The associated symptoms of the chest pain in pediatric patients

Symptom	N = 352
Palpitation	67 (19%)
Dyspnea	44 (12.5%)
Dizziness	27 (7.6%)
Nausea	23 (6.5%)
Cough	16 (4.5%)
Syncope	15 (4.2%)
Weakness/tiredness	12 (3.4%)
Numbness in the hand and arm	12 (3.4%)
Upper respiratory tract symptoms (sore throat, sputum, runny nose)	12 (3.4%)
Abdominal pain	12 (3.4%)
Cough and dyspnea	9 (2.5%)
Sweating	8 (2.2%)
Vomiting	3 (<1%)
Gastric burn	3 (<1%)
Headache	2 (<1%)
Cardiopulmonary resuscitation	1 (<1%)

check ups. In our case, valvular mild aortic stenosis, mild aortic insufficiency, and severe stenotic coronary arteries were detected. Coronary angiography was performed, and it demonstrated high degree stenosis in the left main coronary artery and right coronary artery. The patient underwent coronary bypass surgery under emergency with the left internal mammary artery to left anterior descending artery, saphenous vein graft to circumflex artery and right coronary artery. Due to intra-aortic involvement in such patients, proximal anastomosis was performed where the antegrade cardioplegia cannula was located. The proximal aorta was heavily calcified in this patient, and the proximal part of the right coronary artery

Table 4. The conditions associated with and exacerbating chest pain

	All patients (N = 352)	Cardiac disease (N = 48)
Resting pain	215 (61%)	21 (43.7%)
Increase with exercise	46 (13%)	12 (25%)
Aggravates with breathing	50 (14.2%)	2 (4.1%)
Increase with lying down	8 (2.2%)	6 (12.5%)
Increase with breathing and lying	9 (2.5%)	7 (14.5%)
Increase with moving forward	2 (0.5%)	0
Related with eating	16 (4.5%)	0
Related with anxiety	6 (1.7%)	0

Table 5. Diagnosis and performed surgery in patients with chest pain

	Diagnosis	Surgery
15-year-old girl	Homozygous FH	CABG, LIMA-LAD, Ao-RCA, Ao-OM
16-year-old boy	ALCAPA syndrome	Extrapulmonary tunnel reconstruction
11-year-old boy	Loeys-Dietz Syndrome	Bentall procedure
9-year-old boy	Kawasaki disease	CABG, LIMA-LAD
16-year-old girl	Rheumatic mitral insufficiency	Posterior-pericardial anuloplasty and anterior chordal shortening
17-year-old boy	RCA arising from left coronary ostium	Lateral pulmonary artery translocation

*FH, familial hypercholesterolemia; ALCAPA, anomalous left coronary artery from pulmonary artery, CABG, coronary artery bypass graft; RCA, right coronary artery; Ao, aorta; LIMA, left internal mammary artery; LAD, left anterior descending, OM, obtuse marginalis

graft was combined with circumflex artery graft to avoid creating a new hole in the proximal aorta. After surgery, the patient was taken to the lipid apheresis program with intensive drug treatment. Myocardial revascularization in pediatric patients mostly included Kawasaki disease, during the surgery for congenital heart disease (CHD) or homozygous familial hypercholesterolemia [Komarov 2020].

Despite the good results of percutaneous intervention in the adult patients with familial hypercholesterolemia, coronary artery bypass graft surgery is preferred in pediatric practice [Ungar 2018; Göksel 2009]. Endovascular intervention also can be performed; however, the patient should be carefully assessed, such as growth or age of the patient.

Kawasaki disease is an acute febrile multisystemic disease characterized by the development of vasculitis with frequent coronary aneurysm in 20–40% of cases. It is the main cause of

ischemic heart disease in children [Gu 2009]. The patient in this study previously was diagnosed with KD two years prior to admission, and he had been following with antiaggregant therapy for two years. A thrombosed aneurysm was observed in the proximal left anterior descending artery in computed coronary angiography of the patient (Figure 2C, D). When the patient's left ventricular functions were deteriorated and ischemia was detected in myocardial perfusion scintigraphy with thallium, he was taken into coronary bypass surgery and left internal mammary artery-left anterior descending artery anastomosis was performed.

Acute myocardial infarction may occur in KD, due to thrombus formation within the aneurysm despite antiaggregant and anticoagulant therapy [Okumori 1995]. Surgical treatment is the preferred approach in Kawasaki disease, both to prevent sudden cardiac death and to improve life quality of the patients [Gu 2009; Kitamura 2009]. Widespread calcifications in coronary arteries, low body weight in children, absence of appropriate stent size, and somatic growth are the main limitations of stent implantation. Arterial grafts for coronary artery bypass graft in children with Kawasaki disease showed significantly better functionality than venous grafts in the long-term [Kitamura 2009; Vida 2013]. The most difficult scenario in this patient is coronary aneurysm with no stenosis, so the graft carries a risk for competitive flow and failure. Our patient underwent myocardial perfusion imaging, and it showed ischemia and segmental wall motion defect. Ligation or reduction of the size of the coronary aneurysm are some of the examples in the patient with aneurysm but no apparent stenosis [Okumori 1995; Kitamura 2018].

Anomalous left coronary artery arising from the pulmonary artery (ALCAPA) generally has been reported as an isolated lesion that constitutes 0.023% of congenital heart anomalies and is seen in 1/300,000 live births [Von Kodolitsch 2004; Cankurt 2017]. Myocardial ischemia develops at the territory of the left coronary system, disturbing left ventricular contractility and mitral valve function [Kazmierczak 2013]. An ALCAPA patient may present with weakness, heart failure, angina, exertional dyspnea, sudden cardiac death, or may be diagnosed incidentally upon coronary angiography and in patients with good collateral circulation, symptoms may not be present [Kristensen 2008; Zheng 2010]. A 16-year-old boy was brought to the hospital emergency room by his relatives with cardiac arrest. Cardiopulmonary resuscitation was performed in the emergency room, and hemodynamic stabilization was established. His past medical history was unremarkable.

Electrocardiography showed diffuse ST depression and T wave inversion and troponin level was mildly elevated. Echocardiography revealed that the left main coronary artery was arising from the pulmonary artery and left ventricular systolic dysfunction (Figure 3A). (Figure 3) After right and left heart catheterization, the patient was diagnosed with ALCAPA syndrome (Figure 3B), and surgery was planned. Treatment options of the ALCAPA were surgery with re-implantation of the coronary artery, restoration of the two-coronary circulation with arterial-venous grafts, or intrapulmonary tunnel reconstruction [Murthy 2001]. In our patient, we decided to

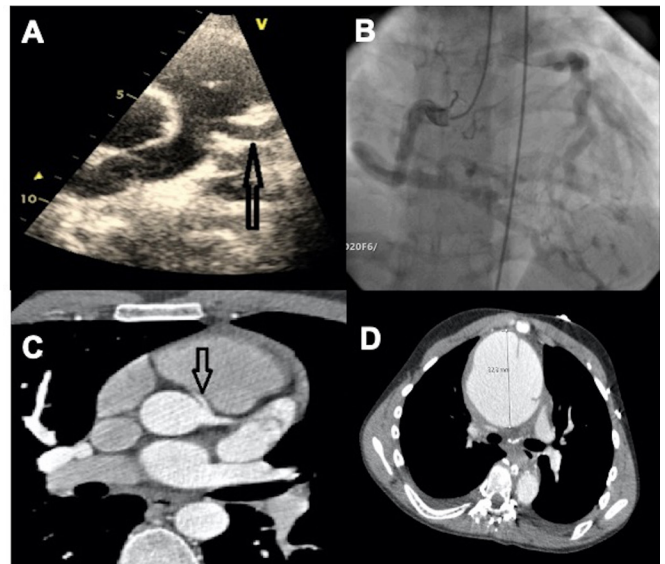


Figure 3. Transthoracic echocardiography showing left main coronary artery (arrow) originated pulmonary artery (A), selective right coronary angiography showing collateral arteries to left coronary system that drain off to the pulmonary artery (B); computed tomography angiography showing the right coronary artery originated from the left coronary sinus and intraarterial course (C); computed tomography angiography showing aortic dilatation and dissection in a patient with Loeys-Dietz Syndrome.

create an extrapulmonary tunnel to connect the abnormal coronary artery to the ascending aorta incompatible with the literature. The pulmonary artery and aortic flaps were used for this tunnel. Therefore, the posterior wall of the tunnel consisted of the pulmonary artery, and the anterior wall consisted of the aorta. We placed the tunnel extrapulmonary, so the pulmonary artery was not exposed to any flexing or twisting. By the way, we avoided the risks associated with intrapulmonary tunneling. Furthermore, extrapulmonary tunnel was formed from autologous tissue, which has the potential to grow and remodel like a normal tissue. The patient was good after surgery and discharged without any problems. The patient has been following up for five years without complications or symptoms.

Anomalous coronary artery arising from the wrong coronary sinus is a rare congenital anomaly, however, certain variants carry a high risk of mortality, with sudden death often being the first manifestation of disease [Gulati 2007]. Surgical correction commonly is recommended when one of these variants is identified, whether symptoms are present. These anomalies mostly are asymptomatic; however, some types of coronary anomalies can cause angina, arrhythmia, syncope, myocardial infarction, and even sudden cardiac death [Frescura 1998]. Intramural course is especially important because coronary artery is squeezed with pulmonary artery and aorta during exercise. Our patient had angina during exercise, and he underwent coronary CT, intra-arterial course of the right coronary artery, which was arising from the left coronary sinus and showed an intraarterial course between

right ventricular outflow track and aorta with no intramural course was detected (Figure 3C). (Figure 3) The lateral pulmonary artery translocation was performed, which previously was defined by Rodefeld et al. in 2001 [Rodefeld 2001]. The main pulmonary artery was carefully transected at the bifurcation level and the arteriotomy widened toward the left pulmonary artery. The right pulmonary artery was reconstructed with a pericardial patch to prevent pulmonary artery stenosis. The main pulmonary artery was translocated left side and re-anastomosed. The patient uneventfully recovered.

A very small percentage – 3.5% – of aortic dissections are seen in patients under 19 years of age. Loeys-Dietz syndrome is an autosomal dominant inherited disease characterized by aortic aneurysm and dissection [Tian 2020; Yoneyama 2021]. The characteristic morphologic findings are hypertelorism, bifid uvula or cleft palate [Singh 2006]. An 11-year-old boy was admitted to our department with chest pain and palpitation for one year. Early diastolic murmur was detected in the left sternal border. Echocardiography showed a dilated ascending aorta and dilatation of aorta. Further evaluation of the patient revealed that the findings were associated with Loeys-Dietz syndrome. He underwent aortic valve replacement and ascending graft (Bentall operation) in another center. Four years after the first surgery, he was admitted to us with aortic dissection and total aortic arch replacement was performed in an emergency setting (Figure 3D). One year later, graft replacement of the descending aorta was performed, due to expanding of the aortic aneurysm and dissection to aortic bifurcation level (Figure 4A). (Figure 4) During follow up of the patient, coronary button pseudoaneurysm and huge left main coronary aneurysm was developed (Figure 4B, C, D). Redo operation was planned, due to risk of rupture. An aggressive surgical approach is recommended to reduce the risk of aortic dissection even in cases with aortic diameter below five centimeters [Williams 2007]. The modified Bentall operation is thought as the standard procedure, however, the valve sparing surgery has become a reliable method for aortic root aneurysms [Fraser 2018; Cameron 2013]. Few case reports were found in literature about the button pseudoaneurysm, this might be excessive normal tissue around the coronary ostia [Fraser 2018].

Acute rheumatic fever could cause mitral insufficiency and may require surgery in some patients [Talwar 2005]. A 16-year-old girl presented with chest pain and exercise intolerance. She had been followed by mitral insufficiency and aortic insufficiency as a sequela of acute rheumatic fever for four years. In our patient, we performed mitral valve surgery due to severe insufficiency, reduced ejection fraction and refractory symptoms of the patient despite drug therapy. Since our patient was an adolescent girl and we wanted to avoid bleeding problems that could be caused by anticoagulant treatment in the future, including pregnancy, we planned a valve repair procedure in the patient. In our patient, we performed valve repair by performing chordal shortening of the anterior leaflet and posterior pericardial annuloplasty. An autogenous pericardium was used for annuloplasty instead of synthetic materials to avoid anticoagulant treatment for a short time. Mitral valve repair is a more accepted method

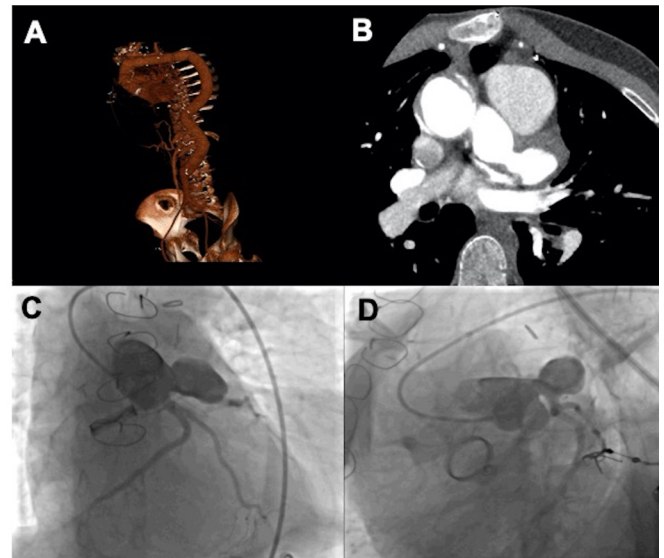


Figure 4. Computed tomography angiography with volume rendering after the surgery showing total aorta replacement (A); computed tomography angiography (B), and selective left coronary angiography (C, D) showing coronary button pseudoaneurysm and left main coronary aneurysm.

compared with the mitral valve replacement technique, especially in children, since it may require additional replacement in the future [Talwar 2005]. Because of the use of anticoagulants, and long-term complications such as thromboembolism, bleeding, rapid degeneration of the prosthetic valve in children and high risk of infective endocarditis, mitral valve replacement is not preferred [Skoularigis 1994]. The valve repair is difficult in rheumatic fever, especially when the anterior leaflet is pathological [Wood 2005]. The intervention should be decided early, depending on the severity of the failure and the degree of valve deterioration, and treatment should be applied before impairment of the left ventricular geometry and functions.

In conclusion, 1.7% of the patients admitted with chest pain to the pediatric emergency department required cardiovascular surgery. Surgical necessity in pediatric patients is quite different, and rare cardiac diseases can be diagnosed with proper examination of the patients.

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