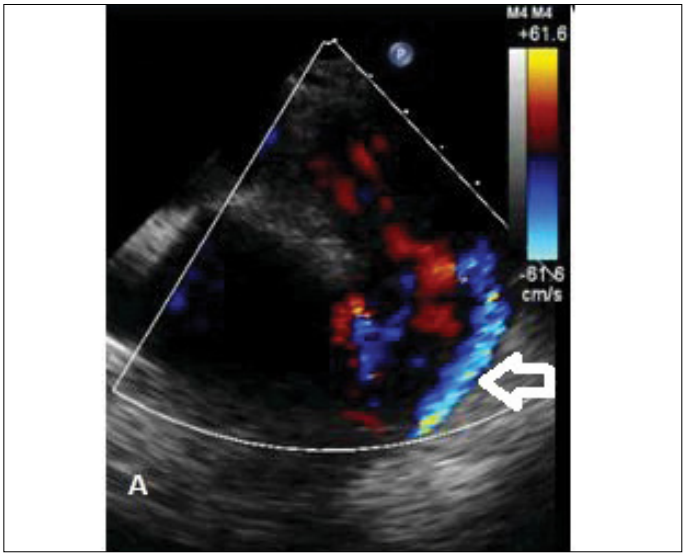

Figure 4. Color Doppler TEE image of a PDA

Table 1. Cases of papillary fibroelastoma associated with congenital heart diseases

Author, year	Age, sex	CHDs	PF location and size, mm
Morishita, 2013	76, M	PDA	AoV, 5
Betigeri, 2011	33, M	AV canal defect (ASD + Cleft mitral)	IVS crest, 20×30
Abad, 2008	60, M	PLSVC, ASD	RA (IAS), 15×20
Watanabe, 1996	64, F	ASD	TV, 11
Current Study			
Patient 1	44, F	ASD	AoV, 9
Patient 2	52, F	ASD, coronary anomaly	AoV, 6
Patient 3	42, F	PDA	AoV, 11

AoV - aortic valve; ASD - atrial septal defect; AV - atrioventricular; CHDs - congenital heart diseases; F - female; IAS - interatrial septum; IVS - interventricular septum; M - male; PDA - patent ductus arteriosus; PF - papillary fibroelastoma; PLSVC - persistent left superior vena cava; RA - right atrium; TV - tricuspid valve.

PDA, which was considered to be caused by suture loosening, and an absence of mass on the aortic valve (Fig. 4). An 8×10-mm Cardiofix device (Starway Medical Technology Inc., Beijing, China) was successfully implanted for PDA. The follow-up course was uneventful. Moderate-to-severe hypertension developed, and nephrectomy was performed a year ago.

Discussion

PFs are uncommon, with an incidence of 7%–8% in all primary cardiac tumors. A majority of PFs occur on the left side of the heart and generally involve the heart valves (1, 2, 7). An association of PF with ASD or other CHDs is rare. To date, four cases of PF associated with CHDs have been reported in the literature (Table 1) (3-6).

In this report, we present a potential new syndrome, which may explain some types of PFs associated with CHDs. To our knowledge, there has been no previous report with direct suggestion of the PF as a more prevalent link of CHDs. Further research on PF associated with CHD syndromes is required with a focus on epidemiology, physiopathological mechanisms, clinical/radiological features, and treatment strategies.

Conclusion

On the basis of the obvious similarities between our cases and those of the other published reports, we propose that a combination of PF and CHDs may represent a recognizable, albeit a rare spectrum of anomalies. We report these cases in the hope that the presence of CHDs will alert the cardiologist to detect a possible PF or vice versa.

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Video 1. Transesophageal echocardiography showed a mass on the aortic valve short axis

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DOI: 10.5152/AnatolJCardiol.2015.6371

Bonsai-induced Kounis Syndrome in a young male patient

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Introduction

The use of cannabis and its synthetic derivative, bonsai, has recently increased, and it has become an important health problem (1). Kounis

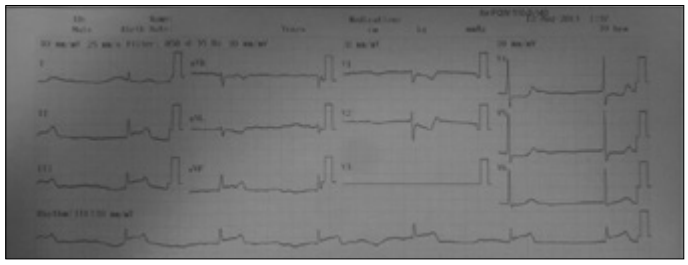

Figure 1. ST elevation in inferior leads on 12-derivation ECG obtained in the emergency department

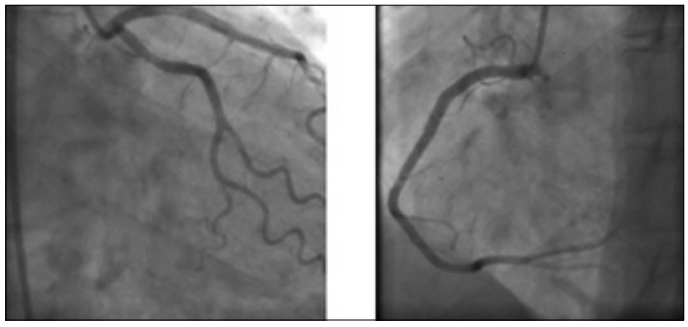

Figure 2. Demonstrating RCA, LMCA, LAD, and LCx on coronary angiography

syndrome develops by the activation of mast cells, and it is an acute coronary syndrome (ACS) related to allergies, hypersensitivity, anaphylaxis, or anaphylactic reactions (2, 3). Bonsai-induced Kounis syndrome has not been reported in literature. The present study presents the case of a 27-year-old patient who arrived at the emergency clinic with chest pain 6 h after the use of bonsai.

Case Report

A 27-year-old male patient arrived at the emergency clinic with sudden-onset retrosternal pain in the left arm, vomiting, and sweating. The chest pain was characterized by pressure and burning and lasted for 6 h. The patient did not have any known atherosclerosis risk factor and reported bonsai use for the first time in his life. He expressed that he had used a great amount of bonsai 1 h before the onset of chest pain. All vital signs of the patient were stable. His electrocardiographic (ECG) investigation revealed mild bradycardia and ST segment elevations in the inferior derivations (D2, D3, and AVF) (Fig. 1). The patient was referred to the coronary intensive care unit after the diagnosis of acute inferior MI was made. Bedside echocardiography revealed inferior and septal hypokinesis. Thrombolytic therapy was planned but was then disregarded as the recently recorded ECG showed ST-segment elevations returning to the isoelectric line. Troponin I value showed a typical increase (4 h)–decrease (24–36 h) (peak value 10.722 ng/mL). Mild leukocytosis and eosinophilia (4.9%) were present. The immunoglobulin E level was high (150 mg/L). The patient was referred to a more advanced center for coronary angiography (CAG). CAG indicated that all coronary arteries were patent (Fig. 2). The fibrinogen and homocysteine levels and antithrombin activity were all within normal ranges. The patient was followed-up for three days without any complications and was then dismissed from the hospital with prescriptions for 100-mg aspirin, 90-mg diltiazem, and 40-mg atorvastatin.

Discussion

To our knowledge, this patient is the first bonsai-induced Kounis syndrome case in literature. Kounis syndrome, in other words allergic MI, has two types depending on the pathophysiology, or the presence of coronary artery disease. In type I, patients exhibit coronary vasospasms induced by allergic mediators such as histamine, thromboxane, and leukotrienes without the presence of atherosclerosis risk factors or coronary artery disease. In type 2, ACS develops due to coronary vasospasms, plaque erosion, or plaque rupture induced by these mediators in patients with atherosclerotic coronary artery disease. Recently, the fact that there are eosinophil and mast cells in the thrombus material excised from some patients in whom stent thrombosis developed after stent implantation with drug release makes us consider hypersensitivity reactions in these patients. This situation is accepted as the type III variant of Kounis Syndrome (4). With these findings, our case is in accordance with the type I variant of Kounis syndrome.

Cardiovascular and psychological problems are frequently reported to be associated with the use of bonsai. The main pathophysiology of Kounis syndrome is the release of many allergic mediators as a result of mast cell activation induced by allergic stimulants. It has been demonstrated in experimental studies that some endogenous cannabinoids suppress inflammation by decreasing mast cell activation via receptors; however, some endogenous cannabinoids trigger mast cell activation independent from receptors (5). In our patient, coronary arteries were revealed to be completely patent, and this may cause us to consider that a coronary vasospasm was the reason that was caused via mediators released by the bonsai-induced activation of mast cells. The main cardiovascular effects are coronary vasoconstriction, increase in the synthesis of tissue factor, thrombocyte activation, dysrhythmia development induced by various mechanisms, and plaque erosion (6, 7). In a patient considered to have Kounis syndrome, in addition to appropriate ACS management, the determination of serum histamine, specific IgE antibodies, and complement proteins and investigation of eosinophilia aid in the diagnosis (2). Leukocyte, eosinophil, and total IgE levels were increased in our patient, but other analyses could not be performed due to technical limitations.

Conclusion

We hoped to emphasize the consideration of the use of bonsai-type synthetic drugs in a young patient with acute MI signs but without any risk factors.

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DOI:10.5152/AnatolJCardiol.2015.6641

Bilateral pulmonary vein stenting for pulmonary vein obstruction after surgical correction of total abnormal pulmonary venous connection

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Introduction

Pulmonary vein stenosis (PVS), either acquired or congenital, is a rare condition that can lead to worsening pulmonary hypertension and cardiac failure in children, and it is frequently lethal. The condition is often progressive and is associated with poor survival (1, 2). Pulmonary vein stenting is an option for acute symptomatic relief and significant improvements in diameter, peak PA pressure/systemic pressure ratio, and trans-stenotic gradient (3). Here we report bilateral PVS in an 11-month-old girl after total anomalous pulmonary venous connection (TAPVC) repair who was successfully treated with bilateral stent implantation.

Case Report

An 8-month-old girl weighing 6 kg was referred to our hospital for surgery. Her initial diagnoses were right atrial isomerism, dextrocardia, unbalanced complete atrioventricular septal defect, double outlet right ventricle, severe pulmonary stenosis, and supracardiac non-obstructive TAPVC. She underwent Glenn anastomosis with TAPVC repair when she was 9 months old. Two months after the surgery, she was referred to our clinic because of cyanosis, respiratory distress, hypoxia, and severe upper extremity and palpebral edema. On admission, she was gasping with bradycardia and severe metabolic acidosis. She was immediately admitted to the pediatric cardiac intensive care; endotracheal intubation and inotropic support were started. Her oxygen saturation level was in the low 70s with 100% oxygen supplement. Her echocardiography revealed pulmonary venous obstruction, Glenn dysfunction, and pulmonary hypertension. Anti-pulmonary hypertensive treatment was added to her treatment. An emergent computed tomography angiography (Fig. 1a) showed severe bilateral pulmonary venous stenosis at the junction of the collector sac and pulmonary vein.

Table 1. Pressure gradients before and after stent implantation

	Pre-intervention	Gradient*	Post-intervention	Gradient
PA	33/13 (mean 26)		26/19 (mean 22)	
RPV	25/23 (mean 24)	11	17/16 (mean 17)	2
LPV	34/26 (mean 27)	14	19/15 (mean 17)	2
CA	mean 13		mean 15	

*Gradients were between the veins and atrium
CA - common atrium; LPV - left pulmonary vein; PA - pulmonary artery; RPV - right pulmonary vein; pressures are in mm Hg.

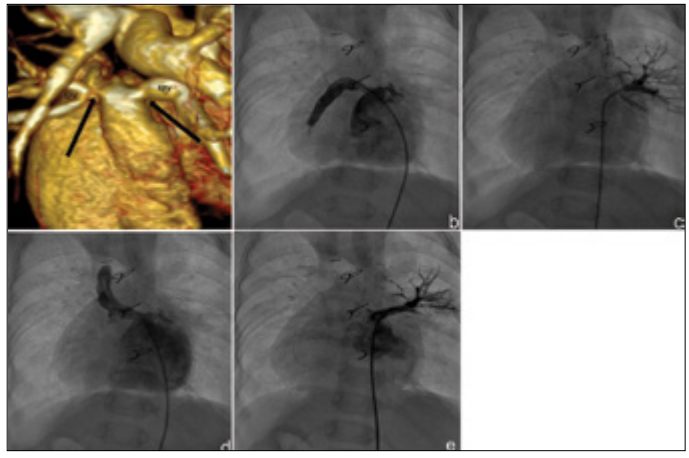

Figure 1. a-e. (a) Posterior volume rendering the multiple detector computed tomography image, (b) posterior–anterior angiographic view of the right pulmonary vein, (c) posterior–anterior angiographic view of the left pulmonary vein, (d) posterior–anterior angiographic view of the right pulmonary vein after stent implantation, and (e) posterior–anterior angiographic view of the left pulmonary vein after stent implantation.
LPV: left pulmonary vein; RPV: right pulmonary vein

Urgent cardiac catheterization for stenting the pulmonary veins was planned. Initially, pressure gradients were gathered (Table 1). Selective right and left pulmonary angiography and direct injection of contrast to the proximal segment of the pulmonary veins showed a narrowing at the junction of the collector sac and pulmonary veins. The narrowest parts were 4 mm and its proximal side was 7 mm on the right pulmonary vein and measurements were 1.5 mm and 4.2 mm, on the left pulmonary vein, respectively (Fig. 1b, c).

Initially, a 7 × 12-mm Palmaz Blue balloon-expandable peripheral stent (Cordis Endovascular, Warren, NJ) was placed across the stenosis on the right pulmonary vein and was dilated until the waist completely disappeared (Fig. 1d, e, Video 1). However, stenting the left pulmonary vein was more complex because it was more stenotic and the left upper and lower pulmonary veins combined together before narrowing. Placing a stent in the lower vein will jail the upper vein or vice versa. After consulting with the surgeons, a 4 × 8-mm Liberte bare coronary stent (Boston Scientific, Natick, MA) was placed across the stenosis. After stent implantation, pressure gradients across the stents dropped to normal levels (Table 1). The patient's oxygen saturation level was elevated to the low 90s. Acetylsalicylic acid, clopidogrel, and standard heparin were initiated after the procedure. She was extubated 3 days after the procedure and was discharged 12 days later. Four months after the procedure, a second catheterization was performed