ZANIMLJIV SLUČAJ KORONARNO-KAMERALNE FISTULE SA ANGINOM PEKTORIS

PRIKAZ SLUČAJA

CASE REPORT

AN INTERESTING CASE OF CORONARY-CAMERAL FISTULA WITH ANGINA PECTORIS

Dejan M. Lazović[1,2], Mladen Kočica[1], Ivana Đurošev[1], Milica Kočica-Karadžić[2,3], Dragan Cvetković[1,2]

[1] Univerzitetski klinički centar Srbije, Klinika za kardiohirurgiju, Beograd, Srbija

[2] Univerzitet u Beogradu, Medicinski fakultet, Beograd, Srbija

[3] Univerzitetski klinički centar Srbije, Centar za anesteziju, Beograd, Srbija

[1] *University Clinical Center of Serbia, Clinic for Cardiac Surgery, Belgrade, Serbia*

[2] *University of Belgrade, Faculty of Medicine, Belgrade, Serbia*

[3] *University Clinical Center of Serbia, Center for Anesthesia, Belgrade, Serbia*

SAŽETAK

Uvod: Koronarno-kameralna fistula (KKF) predstavlja anomalnu vezu između koronarne arterije i srčane komore. Većina KKF-a se otkrije slučajno, tokom angiografske evaluacije koronarnih vaskularnih poremećaja. Prikazujemo slučaj KKF-a sa anginom pektoris.

Prikaz slučaja: Žena, stara 67 godina, imala je bolove u grudima i dispneju pri naporu. Koronarna angiografija je pokazala aterosklerotske lezije u dve glavne koronarne arterije, ali i postojanje komunikacije između tri arterije i šupljine desne komore kroz mnoge male, difuzne fistule. Angiografija je takođe pokazala fistulu između proksimalnog dela leve prednje silazne arterije (prve septalne grane) i desne komore, i između proksimalnog dela desne srčane arterije (akutna marginalna grana) i desne komore. Kod pacijentkinje je postojala indikacija za hiruršku revaskularizaciju miokarda (CABG) i hirurško zatvaranje fistula, te je, na kucajućem srcu, učinjena CABG intervencija, sa dva autovenska grafta – između uzlazne aorte i desne srčane arterije, i između uzlazne aorte i leve prednje silazne arterije, kao i zatvaranje fistula hemoklipsevima i polipropilenskim šavom, sa teflonskim pledžetom.

Zaključak: Hemodinamski beznačajne fistule, koje su klinički neme i nisu povezane sa drugim abnormalnim nalazima, najčešće ne zahtevaju dalje lečenje. Velike, hemodinamski značajne fistule treba ligirati. Ipak, kako manje fistule imaju tendenciju da se uvećavaju sa godinama, preporučuje se i njihovo rano elektivno zatvaranje, u slučajevima kada su one simptomatske, ali i onda kada su asimptomatske, uz prisutan kontinuirani šum ili sistolni šum sa ranom dijastolnom komponentom.

Ključne reči: fistule koronarne arterije, koronarna angiografija

ABSTRACT

Introduction: Coronary-cameral fistula (CCF) is an anomalous connection between a coronary artery and a cardiac chamber. Most CCFs are discovered incidentally during angiographic evaluation of coronary vascular disorders. We report a case of CCF with angina pectoris.

Case report: A 67-year-old woman presented with chest pain and dyspnea upon exertion. Coronary angiography showed atherosclerotic lesions in the two major coronary arteries, but also communication between three arteries and the cavity of the right ventricle (RV) through many small, diffuse fistulas. Angiography also showed a fistula between the proximal left anterior descending artery (LAD) (first septal branch) and the right ventricle, as well as between the proximal right coronary artery (RCA) (acute marginal branch) and the right ventricle. The patient qualified to undergo coronary artery bypass graft surgery (CABG) and surgical closing of the fistulas, which is why we performed, on a beating heart, double vessel revascularization by autovein graft between the ascending aorta and the RCA and between the ascending aorta and the LAD, as well as closing of the fistulas with hemoclips and polypropylene suture, with a teflon pledget.

Conclusion: Hemodynamically insignificant fistulas, which are clinically silent and not associated with other abnormal findings, most commonly do not require further treatment. Large, hemodynamically significant fistulas should be closed by ligation. However, smaller fistulas tend to get larger with age and it is recommended that early elective closure is performed in patients experiencing symptoms or in asymptomatic patients with a continuous murmur or a systolic murmur with an early diastolic component.

Key words: coronary artery fistulas, coronary angiography

Autor za korespondenciju:
Dejan Lazović
Klinika za kardiohirurgiju, Univerzitetski klinički centar Srbije
Pasterova 2, 11000 Beograd, Srbija
Elektronska adresa: lazovic.dejan88@gmail.com

Corresponding author:
Dejan Lazović
Clinic for Cardiac Surgery, University Clinical Center of Serbia
2 Pasterova Street, 11000 Belgrade, Serbia
E-mail: lazovic.dejan88@gmail.com

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UVOD

Koronarno-kameralna fistula (KKF) predstavlja anomalnu vezu između koronarne arterije i srčane komore. Većina KKF-a se otkrije slučajno tokom angiografske evaluacije koronarnih vaskularnih poremećaja. Nekada se smatralo da je koronarno-kameralna fistula, koju je prvi put opisao Krause, 1865. godine, retka anomalija srčanih arterija [1]. Prikazujemo slučaj koronarno-kameralne fistule sa anginom pektoris. Selektivnom koronarnom angiografijom je otkrivena difuzna KKF, koja uključuje desnu srčanu arteriju (engl. *right coronary artery – RCA*) (Slika 1) i levu prednju silaznu arteriju (engl. *left anterior descending artery – LAD*) (Slika 2), koje se prazne u desnu komoru (DK), kao i dvosudovna koronarna bolest.

PRIKAZ BOLESNIKA

Žena stara 67 godina, bez poznatih faktora rizika za srčana oboljenja, imala je bolove u grudima i dispneju pri naporu. Vitalni znaci su bili normalni. Ehokardiografski pregled je pokazao očuvanu ejekcionu frakciju leve komore (LK) – 60%, te odsustvo valvularnog oboljenja, ali je uočena umerena hipertrofija leve komore sa dijastolnom disfunkcijom.

Koronarna angiografija je pokazala aterosklerotske lezije u dve glavne srčane arterije, a otkriveno je i da su tri srčane arterije bile u komunikaciji sa šupljinom desne komore kroz mnoge male, difuzne fistule (Slika 1 i Slika 2), što je za posledicu imalo potpuno zamućenje kontrasta

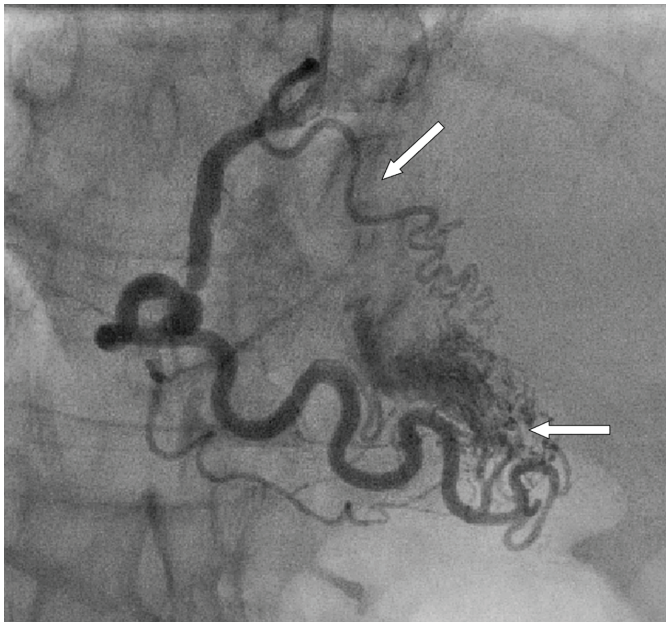
INTRODUCTION

Coronary-cameral fistula (CCF) is an anomalous connection between a coronary artery and a cardiac chamber. Most CCFs are discovered incidentally during angiographic evaluation of coronary vascular disorders. It was previously believed that coronary-cameral fistula, first described by Krause in 1865, is a rare anomaly of the coronary arteries [1]. We present a case of coronary-cameral fistula with angina pectoris. Selective coronary angiography revealed diffuse CCF, which included the right coronary artery (RCA) (Figure 1) and the left anterior descending artery (LAD) (Figure 2), voiding into the right ventricle (RV), as well as double coronary vessel disease.

CASE PRESENTATION

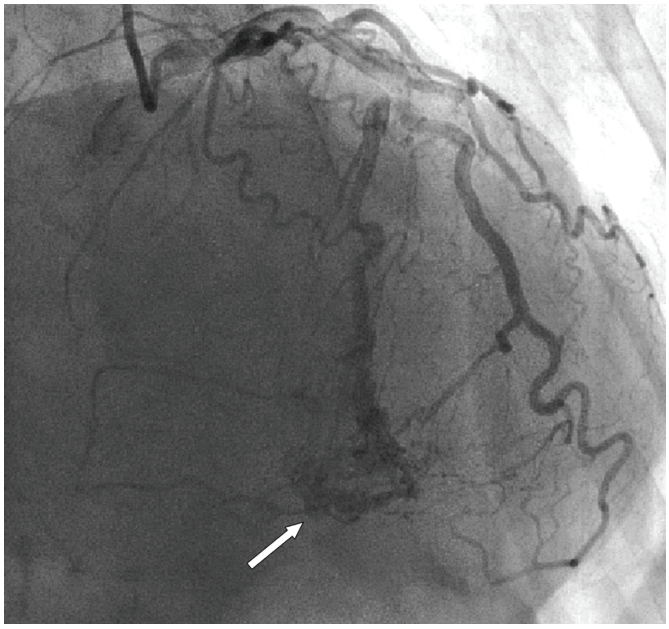
A 67-year-old woman, without known risk factors for coronary disease, presented with chest pain and dyspnea upon exertion. Her vital signs were normal. Echocardiographic examination showed preserved ejection fraction of the left ventricle (LV) – 60%, and absence of valvular heart disease, however moderate hypertrophy of the left ventricle with diastolic dysfunction was observed.

Coronary angiography showed atherosclerotic lesions in the two main coronary arteries, and it was also discovered that three coronary arteries were in communication with the right ventricular cavity through many small, diffuse fistulas (Figure 1 and Figure 2), which resulted in



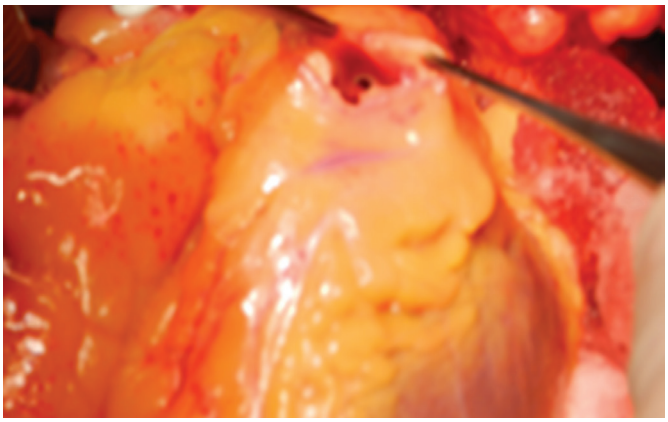
Slika 1. Selektivna koronarna angiografija pokazuje lavirint finih krvnih sudova, koji nastaju iz desne srčane arterije (proksimalno i distalno), i u komunikaciji su sa šupljinom desne komore

Figure 1. Selective coronary angiography showing a maze of fine vessels arising from the right coronary artery (proximal and distal) and communicating with the right ventricular cavity



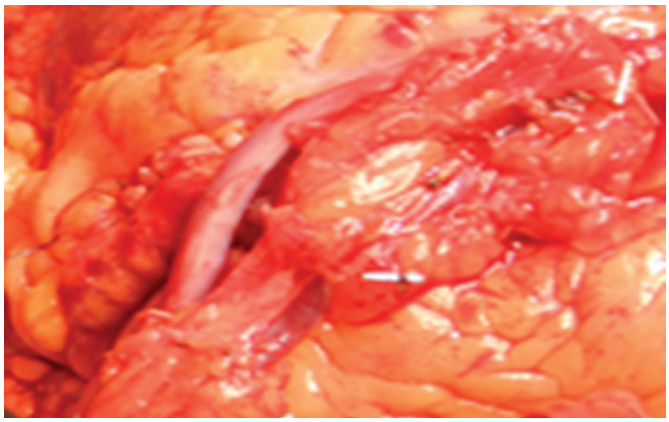
Slika 2. Selektivna koronarna angiografija pokazuje višestruke fistule koje nastaju iz proksimalne leve prednje silazne srčane arterije – prve septalne grane, a koje su u komunikaciji sa šupljinom desne komore

Figure 2. Selective coronary angiography showing multiple fistulas arising from the proximal left anterior descending coronary artery - first septal branch and communicating with the right ventricular cavity



Slika 3. Identifikovan ostijum fistuloznog kanala između RCA-e i DK

Figure 3. The ostium of the fistula between the RCA and the RV



Slika 4. Ligirana i prešivena fistula, bajpas od aorte ka desnoj srčanoj arteriji

Figure 4. Ligated and sutured fistula, by pass from the aorta to the right coronary artery

u desnoj komori. Koronarni sinus je bio normalne veličine. Selektivna koronarografija otkrila je stenozu od 90 – 99% u distalnom delu desne srčane arterije (*RCA*) i stenozu od 80% u kratkom segmentu leve prednje silazne arterije (*LAD*), koja se širila u drugu dijagonalnu granu.

Angiografija je takođe pokazala fistulu između proksimalnog dela leve prednje silazne arterije (prve septalne grane) i desne komore, i između proksimalnog dela desne srčane arterije (akutna marginalna grana) i desne komore.

Transtorakalni, dvodimenzionalni ehokardiogram pokazao je normalne prečnike srčanih komora. Stablo plućne arterije bilo je prepunjeno krvlju. Sistolni pritisak desne komore bio je umereno povećan – do vrednosti od 32 mmHg. Kolor dopler ehokardiografski pregled ukazao je na prisustvo fistule srčane arterije. Pacijentkinja nije bila prethodno podvrgnuta nekoj invazivnoj kardiološkoj dijagnostičko-terapijskoj proceduri koja bi mogla da se dovede u vezu sa KKF-om, a u porodičnoj anamnezi je negirala postojanje koronarno-kameralne fistule.

Kod pacijentkinje je postojala indikacija za hiruršku revaskularizaciju miokarda (engl. *coronary artery bypass graft surgery – CABG*) i hirurško zatvaranje fistule.

U slučaju naše pacijentkinje, na kucajućem srcu, učinjena je hirurška revaskularizacija miokarda, sa dva autovenska grafta – između uzlazne aorte i desne srčane arterije, te između uzlazne aorte i leve prednje silazne arterije. Fistulozni trakt je osiguran sa dva hemoklipsa i polipropilenskim šavom 4-0 sa teflonskim pledžetom (Slika 3 i Slika 4). Pacijentkinja je otpuštena kući u dobrom kliničkom stanju.

DISKUSIJA

Fistula koronarne arterije predstavlja postojanje abnormalne direktne veze između srčane arterije i srčane komore (koronarno-kameralna fistula) ili vene (koronarna

complete contrast opacification in the right ventricle. The coronary sinus was normal in size. Selective coronarography revealed a 90 – 99% stenosis in the distal segment of the right coronary artery (RCA) and an 80% stenosis in the short segment of the left anterior descending artery *(LAD)*, spreading into the other, diagonal branch.

Angiography also showed a fistula between the proximal left anterior descending artery (LAD) (first septal branch) and the right ventricle, as well as between the proximal right coronary artery (RCA) (acute marginal branch) and the right ventricle.

Two-dimensional transthoracic echocardiography showed normal diameters of the cardiac ventricles. The trunk of the pulmonary artery was overflown with blood. The systolic pressure of the right ventricle was moderately elevated – up to the value of 32 mmHg. Color doppler echocardiographic examination indicated the presence of coronary artery fistula. The patient had not previously undergone any invasive cardiological diagnostic or therapeutic procedure that could be connected with CCF, and in her family anamnesis, she negated the existence of coronary-cameral fistula.

The patient's condition indicated the need for surgical revascularization of the myocardium (coronary artery bypass graft surgery – CABG) and surgical closing of the fistula.

In the case of our patient, we performed, on a beating heart, double vessel revascularization by autovein graft between the ascending aorta and the RCA, and between the ascending aorta and the LAD. The fistulas were secured with two hemoclips and polypropylene suture 4-0, with a teflon pledget (Figure 3 and Figure 4). The patient was discharged from hospital in good general condition.

DISCUSSION

Coronary artery fistula is an abnormal direct connection between a coronary artery and a cardiac chamber (coronary-cameral fistula) or vein (coronary arteriovenous

arteriovenska fistula). Fistule srčane arterije se javljaju sa incidencijom od 0,1%. Retko, mogu izazvati ishemiju miokarda, usled sindroma koronarne krađe, srčane insuficijencije ili spontane intraperikardne rupture [2]. Šezdeset procenata ovih fistula nastaje iz desne srčane arterije, a 90% se završava na desnoj strani srca. Koronarno-kameralne fistule, iz sve tri glavne srčane arterije, a koje se završavaju u desnoj komori, retka su pojava.

Koronarno-kameralne fistule su najčešće urođene i mogu biti u vezi sa normalno prisutnim Tebezijevim venama, koje dreniraju krv srčanih arterija nakon njenog prolaska kroz kapilare u srčane komore [3]. One obično nemaju klinički značaj niti daju kliničke simptome i znake. U zavisnosti od veličine i lokalizacije fistula, u nekim slučajevima, mogu se izvesti epikardijalno i endokardijalno hirurško ligiranje ili perkutana endoluminalna procedura (embolizacija). Intervencija je teška ili nemoguća kada su fistule difuzne. Stoga, uprkos ishemiji, kod naše pacijentkinje, takva intervencija nije razmatrana [4].

Jamanaka i Hobs su pronašli fistule malih srčanih arterija kod 0,18% od 126.595 pacijenata podvrgnutih kateterizaciji srca [2]. Fistule ovog tipa obično nastaju iz jedne grane srčane arterije i dreniraju se u jednu od srčanih komora ili u jedan krvni sud, i najčešće nisu povezane sa kliničkim simptomima i znacima [6]. Ipak, velike fistule sa značajnim šantom daju znake kao što je prisustvo kontinuiranog šuma i simptome poput dispneje pri naporu, intolerancije fizičke aktivnosti ili kongestivne srčane insuficijencije, i tada ih treba hirurški korigovati [5,6,7]. Glavna mesta nastanka fistula su desna srčana arterija (55%), leva srčana arterija (35%) i obe srčane arterije (5%). Glavna mesta drenaže fistula su desna komora (40%), desna pretkomora (26%), plućne arterije (17%) i ređe gornja šuplja vena ili koronarni sinus, a najređe leva pretkomora i leva komora [8].

Sukob interesa: Nije prijavljen.

fistula). Coronary artery fistulas occur with an incidence of 0.1%. Rarely, they may cause myocardial ischemia, due to coronary steal syndrome, cardiac insufficiency, or spontaneous intrapericardial rupture [2]. Sixty percent of these fistulas originate from the right coronary artery, while 90% end on the right side of the heart. Coronary-cameral fistulas, from all three major coronary arteries, which end in the right ventricle, are a rare occurrence.

Coronary-cameral fistulas are most commonly congenital and may be connected to the normally occurring Thebesian veins, which drain blood from the coronary arteries after it passes through the capillaries into the cardiac ventricles [3]. They are usually clinically insignificant, and do not result in clinical symptoms and signs. Depending on the size and localization of the fistulas, in some cases, surgical ligation, epicardially or endocardially, or a percutaneous endoluminal procedure (embolization) may be performed. The procedure is difficult or impossible when the fistulas are diffuse. Therefore, despite ischemia, such a procedure was not contemplated in our patient [4].

Yamanaka and Hobbs found fistulas of small coronary arteries in 0.18% of 126,595 patients who had undergone cardiac catheterization [2]. Fistulas of this type usually develop from one coronary artery branch and drain into one of the cardiac ventricles or into one blood vessel, and are most commonly not associated with clinical signs and symptoms [6]. Nevertheless, large fistulas with a significant shunt result in signs such as the presence of a persistent murmur and symptoms such as dyspnea upon exertion, exercise intolerance, or congestive heart failure, which is when surgical treatment is necessary [5,6,7]. The main sites of fistula development are the right coronary artery (55%), the left coronary artery (35%), and both coronary arteries (5%). The main sites where fistulas most commonly drain are the right ventricle (40%), the right atrium (26%), pulmonary arteries (17%), less frequently the superior vena cava or the coronary sinus, and least frequently the left atrium and the left ventricle [8].

Conflict of interest: None declared.

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