

Spontaneous closure of an iatrogenic coronary artery fistula during recanalization of a chronic total occlusion lesion

A case report

Xue Yu, MD, MSc, Xinyue Wang, MD, Ruisheng Zhang, MD, Feng Xu, MD, Fusui Ji, MD*

Abstract

Rationale: Coronary perforation leading to fistula directed to the right ventricle is a rare complication of percutaneous coronary intervention (PCI). The reported outcomes vary from a stable state to rapid deterioration.

Patient concerns: An 86-year-old man was diagnosed with non-ST elevation myocardial infarction, and arranged to PCI procedure for the chronic total occluded right coronary artery (RCA) after coronary angiography. The guide wire went through the occluded lesion and got to the distal part of the suspected “post lateral artery”, which later proved to be in the right ventricle (RV). After dilating with a 2.0 mm balloon, large amount of contrast medium leaked out from the lesion; therefore, we suspected a perforation into the pericardium.

Intervention: Protamine was intravenously injected to convert the effect of heparin and the 2.0 mm balloon in diameter was dilated for about 1 h to obstruct the ejected blood flow shunting into the pericardium, but the leakage persisted. Nevertheless, the patient remained stable, and we were unable to detect an effusion in the pericardium.

Diagnosis: By analyzing the angiogram and echocardiogram, we found that the contrast did not leak into the pericardium, but into the right ventricle (RV) chamber. An iatrogenic coronary artery fistula (ICAF) from the RCA to the RV was confirmed. We thus terminated the procedure.

Outcomes: Coronary computed tomography (CT) angiography was performed 2 days after the PCI and no abnormal shunt was found. There was no abnormal Doppler signal in the RV, either. The patient was soon discharged, and there have been no complaints of discomfort during the 10-month follow-up.

Lessons: ICAFs from coronary to the RV always have favorable outcomes. Even like the one in this case that caused medium leakage could seal spontaneously without any additional management. Echocardiography or coronary CT angiography could be chosen as imaging options to follow-up ICAFs.

Abbreviations: CCFs = coronary cameral fistulas, CT = computed tomography, CTO = chronic total occlusion, ECG = electrocardiography, ICAF = iatrogenic coronary artery fistula, ICD = implantable cardioverter defibrillator, LAD = left anterior descending coronary artery, LCX = circumflex artery, NSTEMI = non-ST elevation myocardial infarction, PCI = percutaneous coronary intervention, PLA = post lateral artery, RCA = right coronary artery, RV = right ventricle.

Keywords: chronic total occlusion, complications, fistula, percutaneous coronary intervention

Editor: N/A.

The patient has provided informed consent for publication of this case.

All authors declare that they have no conflict of interest.

Supplemental Digital Content is available for this article.

Department of Cardiology, Beijing Hospital, National Center of Gerontology, Beijing, China.

* Correspondence: Fusui Ji, Beijing Hospital, National Center of Gerontology, No. 1 Dongdan Dahua Road, Dongcheng District, Beijing 100730, China (e-mail: jifusuivip@126.com).

Copyright © 2019 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

Medicine (2019) 98:3(e14068)

Received: 14 August 2018 / Received in final form: 10 December 2018 /

Accepted: 18 December 2018

<http://dx.doi.org/10.1097/MD.0000000000014068>

1. Introduction

Percutaneous coronary intervention (PCI) is a commonly used procedure to treat coronary artery disease. Iatrogenic coronary artery fistula (ICAF) is a rarely reported complication.^[1] An ICAF is an abnormal communication through which coronary artery blood is shunted into a cardiac chamber or other vascular structure without first passing through the myocardial capillary bed. The treatment strategy of ICAF is not well established and mainly depends on the patient's hemodynamic status.^[2,3] Here we report on a case in which the ICAF spontaneously resolved during intervention for a chronic total occlusion (CTO) lesion.

2. Case report

An 86-year-old male patient underwent coronary angiography for non-ST elevation myocardial infarction (NSTEMI) with inferior electrocardiography (ECG) changes. The angiogram

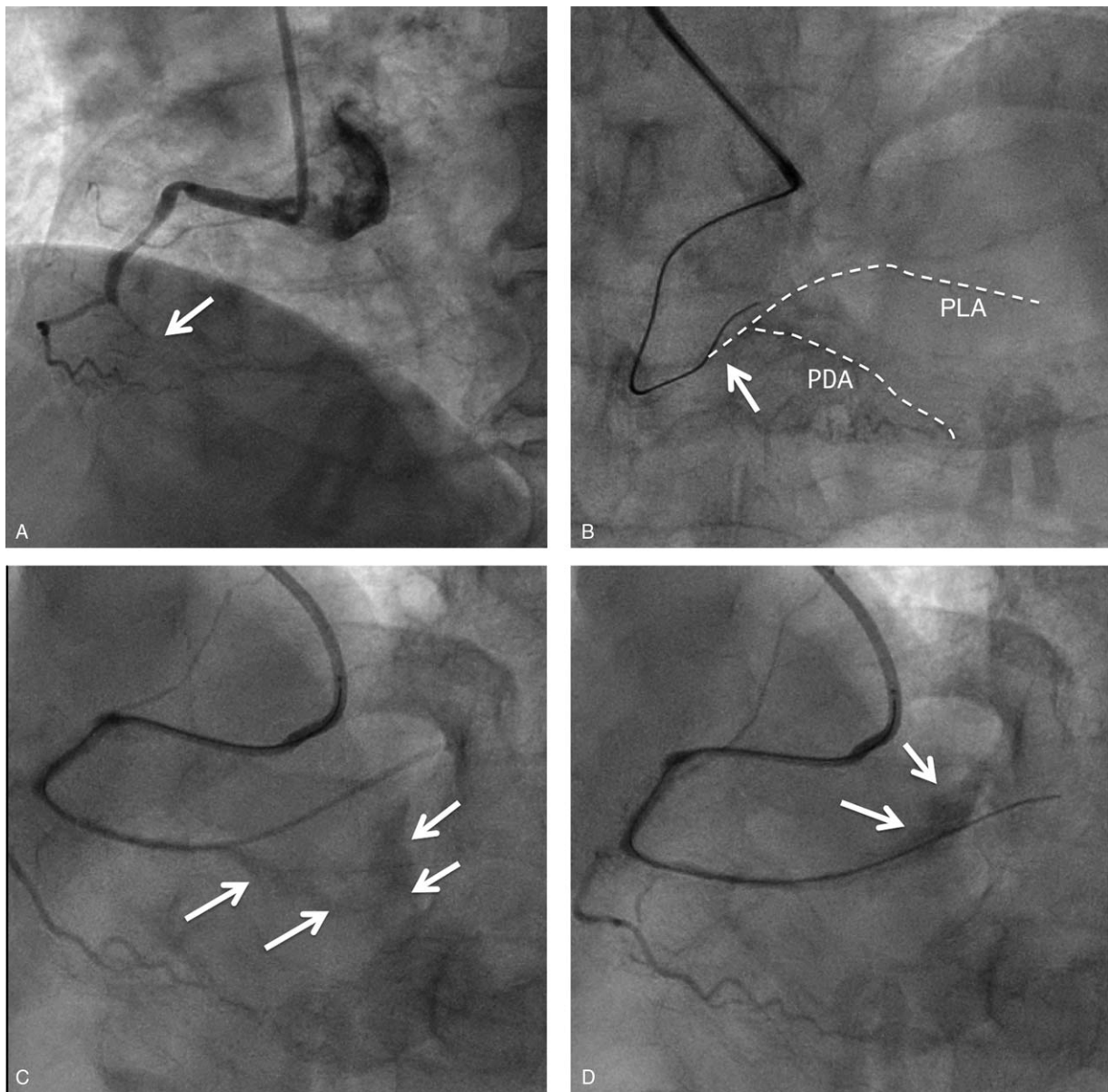


Figure 1. A: Coronary angiogram showing a total occlusion lesion from the second bend of the RCA. B: The guidewire had been wrongly placed in the RV. The arrow indicates the entry site from the RCA to RV. The white dotted lines indicate the true direction of the PDA and PLA according to the calcium shadows. A jet of contrast (arrows) leaking out from the distal CTO lesion immediately after the 2.0mm dilated balloon both at systolic phase (C) and diastolic phase (D). Dynamic images can be seen in Video 1, <http://links.lww.com/MD/C752>. CTO=chronic total occlusion, PDA=posterior descending artery, PLA=posterior lateral artery, RCA=right coronary artery, RV=right ventricle.

showed that the left anterior descending coronary artery (LAD) was patent, and that the distal part of the circumflex artery (LCX) had a CTO lesion and had abundant bridge collateral circulation. The right coronary artery (RCA) was also totally occluded from the second bend and was supplied through a few collateral paths from the LAD (Fig. 1A). We planned to tunnel the RCA lesion first. Since the collateral circulation from the LAD was too small to navigate the direction of the distal RCA and also considering the poor renal function of our old patient, we did not perform a contralateral angiography. A Medtronic 6F SAL 1.0 guiding catheter was engaged through the ostium of the RCA. The guidewire went through the occluded lesion and got to the distal part of the “post lateral artery” (PLA), which later proved to be in the right ventricle (RV) chamber (Fig. 1B). However, the

microcatheter could not pass through the lesion. Therefore, a 1.25mm balloon (The number indicate the diameter of the balloon) was used, which successfully crossed the lesion and dilated the occluded lesion from the PLA to the distal of the RCA, but we were unable to detect any blood flow. Then a 2.0mm balloon was inserted and dilated the above lesions again. The followed cine showed that the contrast had flushed out from the distal RCA lesion (Fig. 1C and D, Video 1, <http://links.lww.com/MD/C752>). The coronary perforation was documented and classified as Ellis type III^[4] in the RCA. A 2.0mm balloon was immediately inflated in proximity to the perforation site to obstruct blood flow, whilst the pericardiocentesis kit was prepared to be ready for use. To reverse heparin, 20 mg of protamine was intravenously injected. Prolonged obstruction

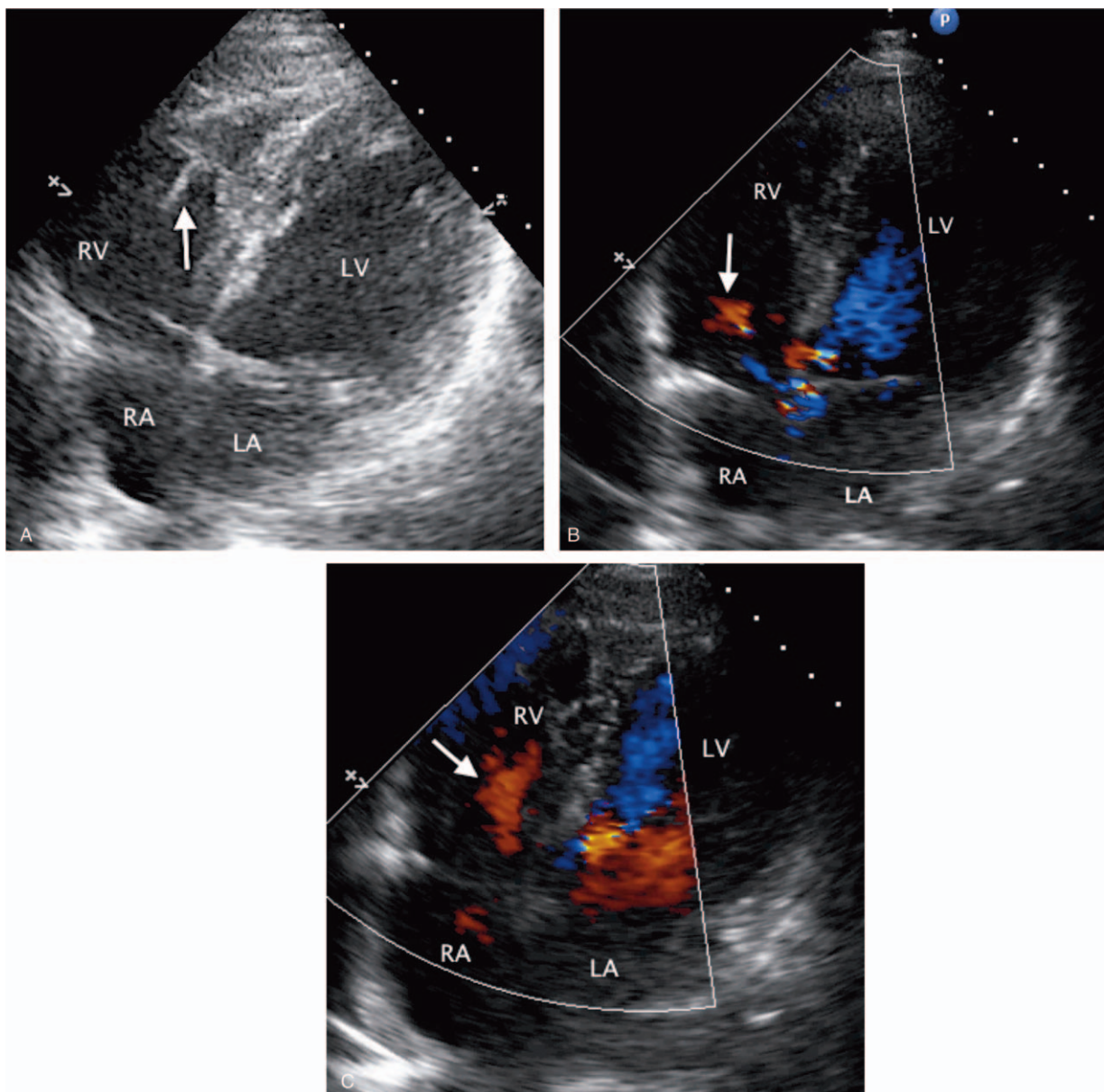


Figure 2. A: An apical four chamber view showing the guidewire signal in the RV chamber (arrow) and the suspected fistulous flow (arrows) in the diastole phase (B) and systole phase (C) in the tricuspid valve region of the RV. It is of importance to note that the flow is more pronounced in the diastole phase and less, but continuous, through the systole phase. Dynamic images can be found in Video 2, <http://links.lww.com/MD/C755>. LA=left atrium, LV=left ventricle, RA=right atrium, RV=right ventricle.

with the 2.0 mm balloon at 8 ATM was performed intermittently for about 60 min, but the leakage persisted. The angiogram showed that the contrast was clearing rapidly with no residual pericardial staining. Echocardiography was repeated several times, but we were unable to detect signs of pericardial effusion nor cardiac tamponade. A very strong echo signal was detected in the RV from the guidewire (Fig. 2A), which confirmed the existence of the perforation. There was an abnormal Doppler flow detected in the RV, which was not seen before PCI and was suspected come from the fistula. The Doppler flow seemed more pronounced in the diastole phase and less, but continuous, through the systole phase (Fig. 2B and C, Video 2, <http://links.lww.com/MD/C755>). The patient remained hemodynamically stable and displayed no symptoms for about 30 min of observation after releasing of the balloon. A careful review of

several angiograms that were obtained at different projections allowed us to determine that the contrast medium did not persist in the myocardium or in the pericardium, but was quickly washed out through the pulmonary artery, suggesting an ICAF from the RCA to the RV. We terminated the procedure and sent the patient to the coronary care unit (CCU) for intensive monitoring. The cardiac enzyme level was within the normal range, and we were unable to detect a pericardial effusion or wall motion abnormalities through echocardiogram. There was no abnormal Doppler signal in the RV either. Two days after the procedure, a coronary computed tomography angiography (CTA) was performed, with the intention to find the fistula located from the RCA to the RV, but no abnormal shunt was observed. The patient was soon discharged and displayed no chest discomfort during the 10 months of follow-up.

3. Discussion

Coronary perforation is a potentially lethal complication during PCI, which may lead to tamponade, myocardial infarction, emergency surgical intervention or death. The incidence of iatrogenic coronary perforation after PCI has been reported to be between 0.1% and 0.58%.^[5-7] However, if plaque debulking techniques were used during the PCI procedure, such as atherectomy, rota-ablation and excimer laser angioplasty, then the incidence of iatrogenic coronary perforations would increase to 0.5 to 3.0%.^[8] Additional perforation risk factors include using a hydrophilic guidewire, oversized balloons or stents,^[9] a high pressure dilation, diffuse vessel disease, chronic total occlusion, old age, and being female.

Coronary perforation that leads to a fistula directed to the RV is a rare complication of PCI.^[10] It was first reported by Meng et al in 1985 that an intracardiac perforation occurred at the LAD to the RV during PCI.^[11] Said et al reviewed PubMed publications focusing on congenital or acquired coronary cameral fistulas (CCFs) between 2000 and 2010.^[11] Publications describing patients with coronary-vascular fistulas were excluded. A total of 243 adult patients were selected who had congenital (n=159/243, 65%) and acquired (n=84/243, 35%) CCFs. Among the acquired-type group, there were 65 patients who had the iatrogenic type of CCFs, and in 15 patients (15/84, 18%) the CCFs were brought about by non-surgical procedures, such as PCI, implanting pacing and implantable-cardioverter defibrillator (ICD) leads and radio-frequency ablation.

Ellis type III perforations were associated with high rates of tamponade, urgent bypass surgery and death. However, type III perforations with cavity spilling, here we call these ICAF, were associated with relatively good outcomes, but the conventional management is still being debated on.^[12] Spontaneous closure has been reported on for congenital CCFs, and it has also been demonstrated in acquired CCFs. In another study, 14 out of 176 heart transplant patients who underwent endomyocardial biopsy experienced ICAF. Fifty-two percent arose from the RCA, 43% from the LAD, and 5% from the LCX. All drained into the RV. No complications of coronary artery fistula developed during a mean follow-up of 28 months. The lack of untoward events, the generally small size of the shunts and the occurrence of spontaneous resolution favor nonsurgical management.^[2] Iatrogenic left coronary artery fistulas were detected in 9 out of 40 study patients with hypertrophic cardiomyopathy, who underwent surgical myectomy. In these cases, only 1 fistula did not heal spontaneously and appeared with ischemic symptoms that needed therapeutic intervention.^[3]

As for ICAF due to PCI, reported cases show a variety of outcomes from stable state to rapid deterioration. Several cases of the spontaneous closure of ICAF complicating PCI have been reported; the reasons for this include guidewire injury^[13-15] and balloon dilatation.^[6,16] In this present case, the shunt was seemed to be of medium size, but the post-procedure echocardiogram and CTA examinations showed no signs of fistulas from the RCA to the RV, which implies that it might have healed spontaneously soon after PCI. This might be due to the weak antegrade blood flow caused by the diseased RCA and the early administration of protamine to reverse the effect of heparin. In other cases, serious complications have been described, such as medium or large fistulas that are associated with volume overload and/or distal myocardial flow impairment, possibly leading to myocardial ischemia.^[17] Ruggieri et al described a case of iatrogenic fistula

from the LAD to the RV caused by excessive post-dilatation after stent implantation at the LAD. They immediately deployed a cover stent and successfully sealed the coronary fistula, but that caused a further complication, coronary thrombosis. The pharmacological management of this is cumbersome in the setting of a coronary perforation.^[18] Previous reports and our experience from this case suggest that small- or even medium-size fistulas may not need to be closed immediately if the patient is hemodynamically stable. Close observation and re-evaluation by echocardiography or CTA are helpful to choose appropriate management strategies, avoiding unnecessary cover stent implantation or coil embolization therapy. If the large iatrogenic coronary-cameral shunt causes myocardial ischemia symptom or heart failure, it should be treated either with percutaneous transcatheter embolization techniques^[19-21] or when the anatomy is not amenable, the surgical ligation method could be utilized.

Provided that most ICAFs occur during PCI were due to coronary perforation, why the blood (contrast) all rush into the chamber in steady of into the pericardial sac. Is it possible that the fistulas already existed there, and that the balloon dilation or guide wire damage that made the fistulas apparent? Said et al analyzed 7 patients with acquired CCF to assess the possible involvement of coronary atherosclerosis in the pathogenesis.^[22] None of the patients underwent therapeutic endovascular procedures before the detection of the fistulas with coronary angiography. They found that 5 out of the 7 patients had atherosclerotic changes and 2 patients were associated with myocardial infarction (MI). CCFs complicating MI has previously been described in the literature.^[23,24] The fistulas had formed as a result of the transmural MI facilitated by the occurrence of soft necrotic myocardial tissue in the infarction region. In the 5 patients associated with severe atherosclerosis, the most likely mechanisms for the formation of the CCFs are: (1) aberrant development of collateral vessels, that is, collaterals that “lost their way” or direction and terminated elsewhere (inadvertently into a cardiac chamber), rather than at the target myocardial region; (2) dilatation of arteriovenous channels that may permit enough shunting of blood to be visualized during coronary angiography.^[25,26]

According to these mechanisms, we surmise that in our case there were already hidden fistulas from the RCA to the RV around the CTO lesion. The guidewire was mistakenly led into the RV through the tunnel of these “hidden” fistulas, and what we eventually dilated was the tunnel but not the free vessel wall. Thus, no “real” perforation was caused, which might explain why there was no sign of an effusion in the pericardium all along.

4. Conclusion

The appropriate treatment for ICAF should be determined on an individual-specific basis according to the severity of the ICAF. Our case indicates that even medium-size ICAFs may seal spontaneously without additional management. Echocardiography or coronary CT angiography could provide useful information during the follow-up of the ICAF.

Acknowledgments

Authors would like to acknowledge Chenguang Yang, Sheng Jiao, and Bo Xia for patient images recording and analysis.

Author contributions

Conceptualization: Xue Yu, Feng Xu, Fusui Ji.
Data curation: Xinyue Wang, Ruisheng Zhang.
Resources: Ruisheng Zhang.
Supervision: Fusui Ji.
Writing – original draft: Xue Yu.
Writing – review & editing: Xue Yu.
 Xue Yu orcid: 0000-0002-6633-6055.

References

- [1] Meng RL, Harlan JL. Left anterior descending coronary artery right ventricle fistula complicating percutaneous transluminal angioplasty. *J Thorac Cardiovasc Surg* 1985;90:387–90.
- [2] Sandhu JS, Uretsky BF, Zerbe TR, et al. Coronary artery fistula in the heart transplant patient: a potential complication of endomyocardial biopsy. *Circulation* 1989;79:350–6.
- [3] Sgalambro A, Olivotto I, Rossi A, et al. Prevalence and clinical significance of acquired left coronary artery fistulas after surgical myectomy in patients with hypertrophic cardiomyopathy. *J Thorac Cardiovasc Surg* 2010;140:1046–52.
- [4] Ellis SG, Ajluni S, Arnold AZ, et al. Increased coronary perforation in the new device era. Incidence, classification, management, and outcome. *Circulation* 1994;90:2725–30.
- [5] Ajluni SC, Glazier S, Blankenship L, et al. Perforations after percutaneous coronary interventions: clinical, angiographic, and therapeutic observations. *Cathet Cardiovasc Diagn* 1994;32:206–12.
- [6] Gruberg L, Pinnow E, Flood R, et al. Incidence, management and outcome of coronary artery perforation during percutaneous coronary intervention. *Am J Cardiol* 2000;86:680–2. A8.
- [7] Fasseas P, Orford JL, Panetta CJ, et al. Incidence, correlates, management, and clinical outcome of coronary perforation: analysis of 16,298 procedures. *Am Heart J* 2004;147:140–5.
- [8] Ramana RK, Arab D, Joyal D, et al. Coronary artery perforation during percutaneous coronary intervention: incidence and outcomes in the new interventional era. *J Invasive Cardiol* 2005;17:603–5.
- [9] Qiu XB, Liu L, Hou XM, et al. Spontaneous resolution of iatrogenic coronary artery to right ventricle fistula secondary to percutaneous coronary intervention. *Chin Med J* 2013;126:2195–6.
- [10] Korpas D, Acevedo C, Lindsey RL, et al. Left anterior descending coronary artery to right ventricular fistula complicating coronary stenting. *J Invasive Cardiol* 2002;14:41–3.
- [11] Said SA, Schiphorst RH, Derksen R, et al. Coronary-cameral fistulas in adults: acquired types (second of two parts). *World J Cardiol* 2013;5:484–94.
- [12] Anselmino M, Ravera L, Biondi-Zoccai G, et al. Iatrogenic coronary-to-right ventricle fistula: benign outcome irrespective of patency? *Minerva Cardioangiol* 2008;56:365–9.
- [13] Bata IR, MacDonald RG, O'Neill BJ. Coronary artery fistula as a complication of percutaneous transluminal coronary angioplasty. *Can J Cardiol* 1993;9:331–5.
- [14] Youssef M, Schob A, Kessler KM. Iatrogenic coronary septal artery-to-right ventricular fistula complicating percutaneous transluminal coronary angioplasty with spontaneous resolution. *Am Heart J* 1997;133:260–2.
- [15] Lipiec P, Peruga JZ, Krzeminska-Pakula M, et al. Right coronary artery-to-right ventricle fistula complicating percutaneous transluminal angioplasty: case report and review of the literature. *J Am Soc Echocardiogr* 2004;17:280–3.
- [16] Alejandro Diego-Nieto, Carlos Cuellas-Ramón, Armando Pérez de Prado, et al. Iatrogenic coronary-cameral fistula after percutaneous intervention on the right coronary artery. *Rev Esp Cardiol* 2008;61:430–8.
- [17] Oreglia JA, Bruschi G, Klugmann S. Percutaneous treatment of iatrogenic left-anterior descending artery to right ventricle fistula. *Catheter Cardiovasc Interv* 2010;76:975–7.
- [18] Ruggieri B, Zimarino M, Caterina RD. Iatrogenic coronary-right ventricular fistula complicated by coronary thrombosis. *Cor et Vasa* 2014;56:e379–81.
- [19] Ata Firuzi, Mohammad Javad Alemzadeh-Ansari, Hamid Reza Pour-aliakbar. Transcatheter coil embolization of iatrogenic coronary artery-left ventricle fistula after mitral valve replacement. *J Saudi Heart Assoc* 2017;29:148–52.
- [20] Freund JE, Yuko-Jowi C, Freund MW. Transcatheter embolization of a large aneurysm in a congenital coronary cameral fistula from the left coronary artery to the right ventricle. *Catheter Cardiovasc Interv* 2015;85:435–9.
- [21] Julian W, Hobbs C, Kumar S, et al. Late presentation of an iatrogenic circumflex to left atrial fistula closed with a covered stent. *J Interv Cardiol* 2004;17:179–81.
- [22] Said SAM, van de Werf T. Acquired coronary cameral fistulas: are these collaterals losing their destination? *Clin Cardiol* 1999;22:297–302.
- [23] Shimizu M, Yoshio H, In0 H, et al. Histopathologic findings of a left coronary artery fistula to the left ventricle after myocardial infarction. *Cardiol Elder* 1993;1:335–6.
- [24] Somsen GA, Liem KS. Coronary artery-left ventricular fistulas probably due to transmural myocardial infarction: a case report. *Cardiologie* 1994;1:65–7.
- [25] Ray SG, Cowan MD, Kennedy JA. Spontaneous acquired fistula from the right coronary artery to the right ventricular cavity. *Br Heart J* 1992;67:323–4.
- [26] Searcy RA, Stein PD, Gunesan G, et al. Arterio-atrial shunting in coronary atherosclerosis. *Chest* 1971;59:398–401.