Successful transcatheter closure of bilateral multiple coronary arterial fistulae in one session

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Abstract

Coronary artery fistulae represent the most frequent congenital anomalies of the coronary arteries, but multiple bilateral fistulae are a rare condition. Current therapeutic options for symptomatic patients are percutaneous closure and cardiac surgery. Transcatheter closure of fistulae using coils is preferred as an effective and safe alternative to surgery. Here we report the case of a patient with congenital coronary artery fistulae arising from both the left and right coronary arteries draining individually into the right pulmonary artery treated successfully with a transcatheter approach.

Key words: coronary fistulae, transcatheter coil embolization, coronary anomaly.

Introduction

A connection between coronary arteries and a cardiac chamber or great artery is called a coronary artery fistula (CAF) [1]. It is a very rare malformation with an average incidence of 0.3–0.8% during diagnostic coronary angiography [2]. Although most cases are congenital, it can be formed after cardiac trauma or surgery [3]. It usually remains asymptomatic but symptoms such as chest pain, dyspnea, and palpitations can develop due to complications related to fistula which include coronary arterial steal, heart failure, and arrhythmias [2]. Although color Doppler echocardiography can demonstrate abnormal flow originating from coronary arteries to other cardiac structures, in some cases conventional or high-resolution computed tomographic coronary angiography is necessary to visualize the CAF. It is indicated to close the CAF in the presence of a large left-to-right shunt, myocardial ischemia, congestive heart failure due to left ventricular volume overload, and significant hemodynamic compromise [4]. Current therapeutic options are percutaneous transcatheter closure and cardiac surgery [5].

In this paper, we report a case with CAF originating from both right and left coronary arteries draining into the pulmonary artery. Successful percutaneous closure with coil embolization was performed without any complications.

Case report

A 33-year-old man presented with symptoms of anginal chest pain. His medical history was unremarkable. He had no cardiovascular risk factor despite 17 pack years of cigarette smoking. The clinical examination was also unremarkable. A 12-lead electrocardiogram was normal, and the results of an exercise test were equivocal. Transthoracic echocardiography demonstrated normal left ventricular systolic function with normal segmental wall motion and valvular functions. Multidetector computerized tomography coronary angiography revealed patent coronary arteries and multiple fistulae between the right coronary artery (RCA), left circumflex coronary artery (Cx) and the right pulmonary artery (PA). Also, conventional coronary angiography confirmed the multiple coronary to pulmonary artery fistulae (Figure 1). Treatment strategies were discussed with the patient and cardiac surgeons. Since the patient was young, free of atherosclerotic coronary artery disease, and unwilling to undergo surgical closure, the percutaneous approach was chosen.

The procedure was carried out under local anesthesia with sedation and a 6 Fr sheath was inserted in the

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Figure 1. Coronary angiography showing multiple coronary fistulae from the branch of the left circumflex coronary artery (**A**) and the branches of the right coronary artery (**B**)

right femoral artery. The left main coronary artery was cannulated with a 6 Fr extra back-up 3.75 guiding catheter (Launcher, Medtronic Inc, Minneapolis, MN, USA), and was deeply engaged to access the distal Cx. Initially, a hi-torque Whisper LS guidewire (Abbott Laboratories, Abbott Park, IL, USA) was negotiated from the Cx to PA through the fistula. Embolization coils (2.5 mm × 3 cm, 3.0 mm × 4 cm, and 2.0 mm × 4 cm; Barricade Coil System, Blockade Medical, Irvine, CA, USA) were delivered to the distal vessel via a micro catheter (Vasco Plus Braided Microcatheter, Balt Extrusion, Montmorency, France). Control angiography demonstrated occlusion of the fistulous communication between the Cx and PA (Figure 2 A). Then, the right coronary artery was cannulated with a 6 Fr JR 4.0 guiding catheter (Launcher, Medtronic Inc, Minneapolis, MN, USA), and was deeply engaged to access the distal RCA. A 0.014" hydrophilic guidewire (Asahi Sion, Abbott Laboratories, Abbott Park, IL, USA) was used to pass through the fistula between the posterolateral branch and PA. Embolization coils (4.0 mm × 6 cm and 2.0 mm × 4 cm; Barricade Coil System, Blockade Medical, Irvine, CA, USA) were delivered and maintained successful closure of the fistula (Figure 2 B). Last of all, the fistulous connection between the proximal RCA and PA was wired with a hydrophilic guidewire (Whisper LS, Abbott Laboratories, Abbott Park, IL, USA). It was occluded using 3.0 mm × 8 cm, 2.0 mm × 4 cm, and 3.0 mm × 4 cm coils (Barricade Coil System, Blockade Medical, Irvine, CA, USA) (Figure 2 C). There were no major complications such as coil migration, dissection of the feeding vessel or of native coronary arteries, myocardial infarction, death, stroke or infection. The patient's hemodynamics remained stable during the procedure, and there were no electrocardiographic changes indicative of myocardial ischemia. Cardiac enzymes remained at normal

levels after the procedure. The patient reported resolution of his symptoms, and he was discharged on the following day. He has been followed up clinically 3 months after the procedure and he has been asymptomatic.

Discussion

With an estimated prevalence of 1% in patients undergoing coronary angiography, CAF represent the most frequent congenital anomaly of the coronary arteries [6, 7]. Although most cases are solitary, 8% are multiple [4]. They are generally linked to RCA, but bilateral CAF originating from both the left and right coronary arteries, as in our case, are quite uncommon. Symptoms such as chest pain and dyspnea can develop due to the coronary steal phenomenon and heart failure during adulthood. This patient described chest pain with physical exertion suggestive of impaired coronary perfusion as a result of increased blood flow through the CAF [5]. Although the appropriate management of a symptomatic patient is controversial, current therapeutic options are transcatheter closure and cardiac surgery. Transcatheter closure approaches have emerged as a less invasive strategy than surgical correction and are nowadays considered as a valuable alternative to surgical correction with similar effectiveness, morbidity and mortality.

Transcatheter closure can be performed by using embolic coil devices, a Rashkind double umbrella device, Amplatzer duct occluder, detachable balloons, and covered stents [8]. Embolization of CAF using microcoils via either an antegrade or retrograde approach is the preferred method for transcatheter closure nowadays [4]. The selection of a proper closure device and technique is mainly based on the individual anatomic features of the CAF. The assessment of the optimal site for device delivery and estimation of the shunt size were reported as key determining





factors to achieve complete occlusion [4]. The coiled vessel becomes thrombosed and disposal of the shunt provides normal myocardial perfusion. The main limitations of the transcatheter approach are inability to deliver the catheter to the fistula due to excessive tortuosity, inadequate size of the coronary artery and presence of coronary branches at the site of the CAF, which occur in nearly 15% of cases [9]. Therefore, appropriate patient selection before considering percutaneous closure management of a symptomatic CAF is essential. There were three CAF in our cases detected at the diagnostic coronary angiogram prior to the procedure. All of them were completely visualized to determine the adequate coil size required and delivery route. We performed transcatheter closure by an antegrade approach via a 6 Fr femoral access and closed completely all the CAF with microcoils. What made this case different from previous reports in the literature was successful closure of multiple CAF in one session without any periprocedural complications [10–12].

Figure 2. Transcatheter closure of the fistulae with coils. Tortuous fistulae between the side branch of the left circumflex coronary artery and the right pulmonary artery (**A**), posterolateral branch (**B**) and proximal branch (**C**) of the right coronary artery have been closed with coils

Results of percutaneous closure procedures are quite satisfactory; complete occlusion succeeded in 60% of the cases [4]. Periprocedural complications such as arrhythmias, dissection, myocardial infarction, and femoral pseudo-aneurysm can develop in up to one-third of the patients but the mortality rate was not increased compared with surgical closure [4, 5]. Absence of coronary artery disease in our patient supported the feasibility of the procedure without any serious complications. Longterm follow-up after a successful transcatheter closure of CAF demonstrated that most of the patients remained symptom-free and 10–20% had residual or recurrent fistulae. Patients with advanced age and confounding cardiovascular risk factors such as diabetes, hypertension, smoking and dyslipidemia are most likely to fail.

Conclusions

We have demonstrated that multiple CAF could be closed by coil embolization in the same session. This pro-

cedure appears to be a relatively safe and effective alternative to surgical correction especially in young patients without any cardiovascular disorder.

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