Successful Coil Embolization with Assistance of Coronary Stenting in an Adult Patient with a Huge Coronary Arterial-Right Atrial Fistula

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Abstract

A 55 year-old man with huge coronary artery fistula to the right atrium is presented. The Qp/Qs ratio was 1.6. We failed to place simultaneously three interlocking detachable coils for closing the fistula, because the coils were washed out into the right atrium. We deployed the coronary stent at the distal portion of the coronary artery. This procedure enabled us to anchor 7 electrically or interlocking detachable coils and to interrupt the shunt flow. A Doppler flow wire was useful to reveal instantaneously the extent of flow reduction even when contrast angiography was not performed in each procedure.

Key words: coronary artery fistula, coil embolization, trans-catheter closure, coronary stent, Doppler guidewire, coronary flow velocity

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Introduction

Coronary artery fistula may sometimes be asymptomatic and may spontaneously close (1), but it may cause angina, myocardial infarction, endocarditis, atrial fibrillation and heart failure (2). Therefore, in patients with symptoms due to the fistula closure of the fistula or flow reduction is necessary to improve symptoms. Surgical repair of fistula usually results in a good clinical course, but it may cause myocardial infarction or troubles due to anesthesia or cardiopulmonary bypass (3). Catheter-based closure of fistula with various materials has recently been developed; this procedure is less invasive and has a better clinical outcome (4, 5). Among the procedures coil embolization is relatively a easy means to close a fistula (9-11). However, it is difficult to close with coil embolization in some cases with high flow coronary artery fistula, tortuous coronary artery fistula or multiple entry sites (12, 13). Migration of coils was also reported as complications (4, 5, 10). We encountered a case of coronary right atrial fistula with a huge, tortuous coronary artery and high flow velocity, which caused difficulty to anchor the coils in the coronary artery.

Case Report

A 55-year-old man suffered from exertional dyspnea for ten years. He visited our hospital because of severe dyspnea at rest. His body weight was 140 kg and height was 178 cm. His radial arterial pulse was irregular and heart rate was 90 beats/min. The blood pressure was 140/80 mmHg. Heart sound was distant and heart murmur was not evident because of marked obesity. Edema was observed in the lower limbs. His chest X-ray showed an increase in pulmonary vasculature and pleural effusion (Fig. 1). The cardiothoracic ratio was 62%. His ECG showed atrial fibrillation. The left coronary artery was dilated and a mosaic color Doppler signal was evident in the right atrium by echocardiography. The left ventricular diastolic dimension was 70 mm and left atrial dimension was 54 mm. The inferior vena cava dimension was 22 mm. The left ventricular ejection fraction was 49%.

Coronary angiography showed marked dilatation and tortuosity of the circumflex coronary artery, and drainage into the right atrium (Fig. 2a, b). Intracardiac blood gas study revealed 5.88 l/min in pulmonary flow and 3.69 l/min in sys-

Figure 1. Chest X-ray showing cardiomegaly, increased pulmonary vasculature and pleural effusion.

temic flow (the Qp/Qs ratio=1.6). Intracardiac pressure was 8 mmHg in mean right atrial pressure, 25 mmHg in mean pulmonary artery pressure and 16 mmHg in mean pulmonary wedge pressure.

Coil embolization technique:

Two weeks after the routine cardiac catheterization, coil embolization of the fistula was attempted. Heparin of 10,000 units was administered intravenously. We considered that a single coil would not stay in the coronary artery and simultaneous multiple coil placement was necessary for closing the high flow huge coronary artery. Two 8 F AL2 guide catheters (Bright Tip, Cordis, Miami Lakes, FL, USA) were inserted from the bilateral femoral arteries. The two guiding catheters were put to the orifice of the left coronary artery. A 0.014 inch PTCA guidewire (Runthrough, Terumo, Japan) was used for inserting an intra-coronary 25 MHz ultrasound catheter (Intrafocus, Terumo, Tokyo, Japan) or the microcatheters for coil embolization. The proximal portion of the left circumflex coronary artery was 8 mm in diameter by intra-coronary ultrasound. The flow velocity was monitored during the procedure by a Doppler guidewire (Volcano Therapeutics, Rancho Cordova, CA, USA), which was placed in the mid-portion of the circumflex coronary artery. The mean velocity was 92 cm/sec at the baseline. Coronary coil embolization was attempted with the guidance of Doppler guidewire flowmetry, which showed the process of the coronary flow reduction instantaneously. No chest pain and no STT changes were evident by coronary occlusion test with a balloon catheter (Naviballoon, protect D type, Kaneka, Osaka, Japan) in the distal portion of the circumflex coronary artery. However, the occlusion balloon was washed out into the right atrium about 20 seconds after inflation. At first, we attempted to place simultaneously two or three complex helical platinum detachable coils, entangled with each other through three microcatheters (Excelsior

and Renegade, Boston Scientific, Boston, MA, USA, and Transit, Cordis, Miami Lakes, FL, USA), in the circumflex coronary artery. However, the coils stayed only a few seconds in the coronary artery and were easily washed out into the right atrium because of high flow velocity. We could not detach the coils. Therefore, some anchors were necessary to keep the coils in the coronary artery. We planned to place the coronary stent (Driver, Medtronic, Minneapolis, MN, USA) of 4 mm in diameter and 18 mm in length at the proximal site of the bent part of the circumflex coronary artery (Fig. 3). Although the larger stent could stay in the coronary artery, the proximal site of the circumflex coronary artery was also tortuous and so we chose the 4 mm in size and the flexible one. The stent was deployed by balloon inflation pressure of 16 atm and it remained in the distal portion of the posterolateral branch of the circumflex coronary artery. This procedure enabled us to anchor the 2 helical platinum electrically detachable coils (GDC coils, 10 mm/30 cm, 9 mm/30 cm, Target Therapeutics, Bayside Parkway Fremont, CA, USA), and 3 interlocking detachable coils (8 mm/20 cm, 7 mm/20 cm, 6 mm/20 cm, Target Therapeutics, Bayside Parkway Fremont, CA, USA) and 2 platinum electrically detachable fibered-coils (GDC fibered 5 mm/8 cm, 4 mm/5 cm, Target Therapeutics, Bayside Parkway Fremont, CA, USA) one by one from large size to small size at the coronary stent (Fig. 4). The flow velocity of proximal side of the coils by Doppler flowmetry gradually decreased when additional coils were positioned. However, the flow velocity did not reach zero level. As the last coil was placed near the posterolateral branch, left coronary angiogram was performed and it revealed almost an interruption of contrast medium at the coils (Fig. 4, right panel). Since an additional coil embolization might close off the postero-lateral branch, we stopped the procedure. Finally, the mean flow velocity of the proximal side of the coils decreased from 92 cm/sec to 8 cm/sec (Fig. 5). No STT changes occurred during or after the procedure and no myocardial enzymes were elevated after the procedure. No other complications occurred during or after the procedure.

Follow-up

By transthoracic echocardiography 8 months after the procedure, the left ventricular diastolic dimension decreased to 61 mm and the ejection fraction increased to 59%. The inferior vena cava dimension decreased to 10 mm. However, a small residual shunt flow was observed by trans-esophageal Doppler echocardiography. Defibrillation for atrial fibrillation by direct electrical current was attempted, but it failed. However, his congestive heart failure improved and no hemolytic anemia was evident.

Discussion

Coronary artery fistula to the pulmonary artery is often observed by routine coronary angiography and it is usually a small shunt flow and asymptomatic. A coronary artery fis-

Figure 2a, 2b. Left coronary angiography of the right anterior oblique view (2a) and the frontal view (2b) showing the markedly dilated and tortuous circumflex coronary artery and its drainage into the right atrium. The circumflex coronary artery was connected to the right atrium near the coronary sinus. LAD=left anterior descending coronary artery, LCX=left circumflex coronary artery.

Figure 3. Stent deployment at the postero-lateral portion of the left circumflex coronary artery (the left panel) and successful placement of the multiple detachable coils at the stent site (the right panel). The Doppler guidewire was placed at the proximal site of the circumflex coronary artery. The PTCA guidewire was inserted along the tortuous circumflex coronary artery and the stent could be employed at the distal site of the two posterolateral branches. Distal tortuosity of the coronary artery made the stent stay at the aimed position.

tula to the right atrium is rare, but it has sometimes large shunt flow. The present case also had moderate shunt flow and heart failure. Surgical repair of the coronary fistula is effective and may provide a good clinical outcome, but it may be invasive and may sometimes cause severe complications such as myocardial infarction, and has a significant risk of recurrence of the fistula (3). Recently, transcatheter procedure for closure of the fistula has developed. The factors against coil occlusion have been distal fistula, adjacent vessel at risk, need for concomitant distal coronary bypass, large fistula, and young age (3). Among them, the present patient had distal and large fistula and adjacent small posterolateral branch at risk. Coil embolization by the old tech-

nique would be a challenging case. However, the risk of occlusion of the adjacent vessel by coil occlusion was denied by the balloon occlusion test. The operative mortality is 1.4% in the surgical literature and 0% in current transcatheter closure (13). Moreover, the present patient had congestive heart failure. We consulted cardiac surgeons in our hospital. They advised that open heart surgery would not be a first choice. Accordingly, we chose coil embolization in this patient.

The materials for occlusion have been Gel foam (6), polyvinyl alcohol foam (7), detachable balloon (8), Gianturco spring coil (Cook, Trevose, PA, USA), detachable coil (Cook, Trevose, PA, USA) (4), Rashkind double-umbrella

Figure 4. A total of 7 various types of coils were positioned (arrow in the left panel). The left coronary angiography of right anterior oblique view after coil embolization showing interruption of the coronary flow at the coils (the right panel). The obtuse marginal and two posterolateral branches were visible after coil embolization. OM=obtuse marginal branch, PL=posterolateral branch.

Figure 5. Flow velocity pattern with a Doppler guidewire positioned at the proximal site of coils. The mean flow velocity decreased from 92cm/sec (upper panel) to 8cm/sec (lower panel) after coil embolization.

device (USCI, Medtronic, Minneapolis, MN, USA) (4), Amplatzer duct occluder (AGA Medical, Golden Valley, MN, USA) (5) and interlocking detachable coils (Target Therapeutics, Bayside Parkway Fremont, CA, USA) (9-11). Among them, recently interlocking detachable coils have been frequently used for pediatric patients due to the controlled release, convenience and less invasiveness. Both the short- and intermediate-term outcomes after transcatheter coil occlusion are good, while persistent coronary artery dilatation is present (5, 13, 14). But in adult patients, the coronary artery of the fistula is markedly dilated and tortuous, and sometimes has a very high flow velocity. These

factors may make it difficult to achieve transcatheter closure of the fistula. The mean flow velocity in the present case was 92 cm/sec and it was about two-fold that of normal coronary flow velocity. Anchoring the detachable coils in the coronary artery may be difficult in such case. As one of complications of the detachable coil embolization, many cases with migration of coils have been reported (4, 5, 10), while the majority of the migrated coils can be retrieved. Okubo et al reported 4 cases with coil migration among 13 children who underwent coronary embolization (5). In the present patient, the coils were easily washed out from the coronary artery to the right atrium, so we did not release the coils in the coronary artery to avoid coil migration. Gianturco coils have been used in one case report with a high flow coronary artery fistula (12), but the delivery system of the coil is 110 cm in length, so it is difficult to place the distal portion of the adult coronary artery through the usual PTCA guide catheter of 100 cm in length. Dorros et al described that the covered stent placement at the orifice of the fistula resulted in successful closure of fistula (15). This method might be useful in patients with multiple entry sites of fistula. However, in the present case the orifice of the fistula was the orifice of the circumflex coronary artery, so covered stent placement could cause posterior wall myocardial infarction.

Sugimoto et al reported, as a personal communication, that simultaneous placement of two interlocking coils entangled with each other led to successful treatment in a case with huge pulmonary arterio-venous fistula (16). We also tried to anchor three platinum detachable coils simultaneously via two guiding catheters. The electrically detachable coils may be safer than interlocking coils in such a high flow case with risk of migration of the coils (17). However, we could keep them in place for only a few seconds in the aimed coronary artery position and abandoned to release the

coils. Therefore, we decided to deploy a coronary stent to anchor the coils. We chose the stent of 4 mm diameter and 18 mm length. The deployed size of the stent would be about 4.2 mm in the inflation pressure. The size of the stent might be smaller as compared with the coronary size, 8 mm. With the use of the smaller stent, there might be the possibility of stent migration. The larger sized stent might be recommended. However, the size of the stent was maximal among the commercially available coronary stents. We could get a larger one if we would choose a stent for systemic arterial use. However, it would be more rigid. Accordingly, there was the possibility that the larger sized stent for systemic artery use could not pass the proximal tortuous portion of the coronary artery. The flexible coronary stent which we chose could easily pass through the tortuous coronary artery, and stayed in the aimed position of the distal circumflex coronary artery. The deployed stent was more rigid and straight than coils or the occlusion balloon, and the distal coronary artery was too tortuous to pass the stent. This might be the explanation why the stent stayed in the coronary artery. The deployed stent could play a role of anchor of coil embolization. We could place numerous detachable coils from large size to small size without coil migration. Armsby et al described the failure of transcatheter occlusion in such a patient with a tortuous coronary artery fistula (13). Our new method for coil embolization with the assistance of intra-coronary stent deployment may provide the successful result even in such complicated cases.

The coronary flow velocity monitoring by a Doppler guidewire was newly conducted during the procedure. The very high flow velocity (92 cm/sec) was elucidated before starting the coil embolization method, and the fact indicated the difficulty of successful embolization. The flow velocity during coil embolization gradually decreased with the placement of each additional coil. The reduction of flow velocity may be based on the increase in coronary resistance due to coil embolization. In other words, the reduction of the flow velocity instantaneously provided the information about the

effectiveness of coil embolization without the need for contrast angiography. However, the flow velocity did not reach zero level despite multiple coils embolization. The reason why the flow velocity did not reach zero was as the follows: The Doppler guidewire was placed in the proximal site of the implanted multiple coils and it detected not only the shunt flow but also the native coronary flow of the left circumflex coronary artery. As there were obtuse marginal and lateral branches of the circumflex coronary artery proximal to the coils, the coronary flow in the left circumflex coronary remained after coil embolization. The attained mean flow velocity was 8 cm/sec and it might be slower than usual coronary flow velocity. As the coronary diameter of the present case was two- or three- fold that of a normal person, the low flow velocity would be reasonable. Accordingly, the final decision of stopping coil embolization depended upon contrast coronary angiography.

In this patient, heart failure might have been caused not only by coronary artery fistula, but also by atrial fibrillation. Defibrillation by direct electrical current was performed, but it failed to restore sinus rhythm. Despite keeping the atrial fibrillation, the left ventricular dimension decreased and the left ventricular ejection fraction improved, and dilatation of the inferior vena cava disappeared. Exercise capacity increased after the procedure. These findings suggest that the main responsible cause of heart failure may be coronary artery fistula. Moreover, there were no acute or chronic myocardial ischemic episodes due to obstruction of the circumflex coronary artery.

Implication

Coronary stent deployment as a role of anchor of coils may be one of the important new techniques for coil embolization of the fistula with a huge coronary artery and a high flow velocity. Doppler coronary flow monitoring may play an important role in the coil embolization method for coronary artery fistula.

References

1. Griffiths SP, Ellis K, Hordof AJ, Martin E, Levine OR, Gersony WM. Spontaneous complete closure of a congenital coronary artery fistula. *J Am Coll Cardiol* **2**: 1169-1173, 1983.
2. Liberthson RR, Sagar K, Berkoben JP, Weintraub RM, Levine FH. Congenital coronary arteriovenous fistula. Report of 13 patients, review of the literature and delineation of management. *Circulation* **59**: 849-854, 1979.
3. Mavroudis C, Backer CL, Rocchini AP, Muster AJ, Gevitz M. Coronary artery fistulas in infants and children: a surgical review and discussion of coil embolization. *Ann Thorac Surg* **63**: 1235-1242, 1997.
4. Perry SB, Rome J, Keane JF, Baim DS, Lock JE. Transcatheter closure of coronary artery fistulas. *J Am Coll Cardiol* **20**: 205-209, 1992.
5. Okubo M, Nykanen D, Benson LN. Outcomes of transcatheter embolization in the treatment of coronary artery fistulas. *Cathet Cardiovasc Interv* **52**: 510-517, 2001.
6. Reidy JF, Jones ODH, Tynan MJ, Baker EJ, Joseph MC. Embolization procedure in congenital heart disease. *Br Heart J* **54**: 184-192, 1985.
7. Strunk BL, Hieshima GB, Shafton EP. Percutaneous treatment of a coronary arteriovenous malformation with micro-particle embolization. *Cathet Cardiovasc Diagn* **22**: 133-136, 1991.
8. Graeb DA, Morris DC, Ricci DR, Tyers GF. Balloon embolization of iatrogenic aortocoronary arteriovenous fistula. *Cathet Cardiovasc Diagn* **20**: 58-62, 1990.
9. Qureshi SA, Reidy JF, Alwi MB, et al. Use of interlocking detachable coils in embolization of coronary arteriovenous fistulas. *Am J Cardiol* **78**: 110-113, 1996.
10. Ogoh Y, Akagi T, Abe T, Hashino K, Hayabuchi N, Kato H. Successful embolization of coronary arteriovenous fistula using an interlocking detachable coil. *Pediatr Cardiol* **18**: 152-155, 1997.
11. Quek Sc, Wong J, Tay JS, Reidy J, Qureshi SA. Transcatheter embolization of coronary artery fistula with controlled release coils. *J*

- Pediatr Child Health **32**: 542-544, 1996.
12. Tayama E, Akagi T, Shojima T. Transcatheter coil embolization of a high-flow coronary artery fistula using 0.052-inch Gianturco coils: a case report. *Circ J* **67**: 1059-1060, 2003.
 13. Armsby LR, Keane JF, Sherwood MC, Forbess JM, Perry SB, Lock JA. Management of coronary artery fistulae. Patient selection and results of transcatheter closure. *J Am Coll Cardiol* **39**: 1026-1032, 2002.
 14. McMahon CJ, Nihill MR, Kovalchin JP, Mullins E, Grifka RG. Coronary artery fistula: Management and intermediate-term outcome after transcatheter coil occlusion. *Tex Heart Inst J* **28**: 21-25, 2001.
 15. Dorros G, Thota V, Ramireddy K, Jaseph G. Catheter-based techniques for closure of coronary fistulae. *Catheter Cardiovasc Interv* **46**: 143-150, 1999.
 16. Sugimoto K, Tsurusaki M, Taniguchi H, et al. Coil embolization with interlocking detachable coils in a patient with giant pulmonary arteriovenous fistula. Usefulness of dual microcatheter method. *Clinical Frontier No 2*, Boston Scientific (in Japanese).
 17. Tsai CW, Lin TH, Ko CT, Chen MF, Lee YT. Transcatheter embolization of a coronary arteriovenous fistula with a complex, helical-fibered platinum coil. *J Formos Med Assoc* **95**: 558-561, 1996.