Transcatheter closure for baffle leak after Takeuchi repair of anomalous left coronary artery from the pulmonary artery: a case report

Hu Hai-Bo*, Yang Kai, and Pan Xiang-bin

Center of Structural Heart Disease, Fuwai Hospital, Chinese Academy of Medical Sciences and Peking Union Medical College, Beijing 100037, China

Received 17 November 2017; accepted 11 February 2018; online publish-ahead-of-print 29 March 2018

Introduction

Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) is a rare but serious congenital coronary abnormality. Takeuchi repair is an alternative treatment option for this anomaly in certain situations, it is reported that baffle leak is the most common complication after Takeuchi repair, and some of this complication require surgical reoperation.

Case presentation

In this case, a 43-year-old woman diagnosed with ALCAPA underwent Takeuchi procedure 6 months ago, 4 months after the procedure, the patient complained of chest pain on exertion and was confirmed to have baffle leak, and then we treated this complication successfully by percutaneous transcatheter closure.

Discussion

Baffle leak is the most common complication after Takeuchi procedure of ALCAPA, some of them require surgical reoperation. In this case, we introduce a new method, percutaneous transcatheter closure, to treat the baffle leak. To our knowledge, this is the first reported case of transcatheter closure for baffle leak after Takeuchi repair, which may be an alternative treatment option for the baffle leak after Takeuchi repair of this rare congenital coronary anomaly.

Keywords

Case report • Coronary anomaly • ALCAPA • Takeuchi • Baffle leak • Transcatheter closure

Learning points

• Baffle leak is the most common complication after Takeuchi procedure of anomalous origin of the left coronary artery from the pulmonary artery. In this case, we introduce a new method, percutaneous transcatheter closure, to treat the baffle leak, which is an alternative treatment option for the baffle leak and may be useful to clinical and interventional cardiologists who care for patients with structural heart disease.

• The interventional procedure should be very soft and careful to avoid the injury and occlusion of the coronary after the implantation of the device.
reported that baffle leak is the most common complication. Since some of this complication require reoperation, we are going to introduce a new treatment method for the baffle leak and our experience of percutaneous transcatheter closure.

**Timeline**

| Time                       | Events                                                                                           |
|----------------------------|--------------------------------------------------------------------------------------------------|
| 20 years ago               | Chest pain occasionally, the patient didn’t pay much attention to it and no special treatment was obtained |
| 6 months ago               | The patient came to our hospital due to worsening symptoms and was diagnosed with anomalous origin of the left coronary artery from the pulmonary artery and treated with Takeuchi procedure |
| Within 3 months after the Takeuchi repair | No abnormality                                                                                   |
| 4 months after the Takeuchi repair | Chief complaint: chest pain on exertion, Physical examination: Grade 3/6 continuous murmur at left sternal border between the 2nd and 4th rib, Imaging: a small fistula from the intrapulmonary baffle to the main pulmonary artery, Treatment: percutaneous transcatheter closure |
| 3 months after the transcatheter closure | No evident residual shunt was observed                                                            |

**Case presentation**

Twenty years ago, a young woman (23 years old) felt chest pain occasionally when she walked hastily, the pain lasted about 2 min and can relieve itself, so she was admitted to the local hospital and diagnosed with mild-moderate mitral insufficiency (MI). Because of no obvious limitation in physical activity, the patient didn’t pay much attention to it, and no special treatment was obtained. Six months ago, she came to Beijing for further diagnosis due to worsening symptoms, she was diagnosed with congenital heart disease, ALCAPA (see Figure 1), severe MI, and left ventricular enlargement (LVE) and underwent Takeuchi procedure and concomitant mitral valve repair in our hospital. During the operation, we found that the left coronary artery (LCA) arises from the left sinus of pulmonary trunk, and we created an aortopulmonary window above the left aortic sinus and an intrapulmonary tunnel that baffles the aorta to the ostium of the anomalous LCA through the anterior wall of the pulmonary trunk. After the operation, she was followed regularly for 3 months and unaware of any abnormalities. Four months after the Takeuchi repair, she began to complain of chest pain on exertion. Physical examination revealed a Grade 3/6 continuous murmur of puffing character at left sternal border between the 2nd and 4th rib, blood pressure (BP) 110/54 mmHg, heart rate 75 b.p.m. Electrocardiogram showed left ventricle high voltage and abnormal Q waves. Transthoracic echocardiogram (TTE) revealed a small fistula from the intrapulmonary baffle to the main pulmonary artery (MPA), mild mitral regurgitation, left ventricular end-diastolic diameter (LVEDD) 54 mm (57 mm before the Takeuchi repair), and ejection fraction 52%. Cardiac computed tomography angiography (CTA) confirmed a fistulous connection from the posterior wall of the proximal LCA into the anterior wall of MPA, the diameter of the fistula is about 4 mm (see Figure 2A).

After the baffle leak was confirmed, we adopted a new method, percutaneous transcatheter closure, to treat the baffle leak. During the operation, ascending aortic angiography in the LAO view demonstrated contrast leakage from the LCA into the MPA (see Figure 3A and B), then we used a ventricular septal defect (VSD) occluder (Starway Medical Technology Inc., Beijing, China) to close the baffle leak through 6 Fr transferring sheath, the VSD occluder we used is a self-expandable, double disc implant device made from a Nitinol wire mesh, the two discs are linked together by a short connecting waist, and the device waist in diameter is 6 mm. Selective coronary angiography of the left main coronary artery in the LAO view demonstrated no contrast leakage after implantation of the device (see Figures 2B and 3C). Blood pressure increased to 115/68 mmHg. The pulmonary artery pressure (PAP) decreased from 46/22 (mean: 29) mmHg to 31/14 (mean: 19) mmHg. The Qp/Qs decreased from 1.52 to 1.05. Twenty-four hours after the interventional treatment, TTE re-examination showed no evident residual shunt and LVEDD decreased from 54 mm to 50 mm. The patient’s symptom improved significantly before hospital discharge, and TTE re-examination of 3-month follow-up showed no evident residual shunt.

**Discussion**

Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) is a rare but serious congenital heart disease and requires surgical repairs once ALCAPA is diagnosed. Takeuchi repair is an alternative treatment option for this anomaly in certain situations, although there is some controversy about this procedure. It was first described in 1979, which consists of using a flap derived from the anterior wall of the pulmonary trunk to create an internal tunnel in the main pulmonary trunk between a surgically created aortopulmonary window and the left coronary ostium, the opening in the pulmonary trunk is patched with pericardium. Reported complications of Takeuchi repair mainly include baffle leak and supravalvular pulmonary stenosis, and the baffle leak is the most common complication (27%). We reviewed related articles in recent 20 years and summarized some information about the baffle leak after Takeuchi repair of ALCAPA (see Table 1). It shows that the incidence rate of baffle leak is about 27.8%, which is similar to the study by Ginde et al., and 9.7% of patients received surgical reoperation. In this case, the patient was confirmed to have baffle leak 4 months after the Takeuchi repair, and the diameter of the fistula was about 4 mm, which was likely to explain the reason why the patient felt chest pain on exertion. In addition to the patient’s
symptom, this complication may result in certain sequelae if untreated, such as chronic myocardial ischaemia and angina, myocardial infarction, heart failure, pulmonary hypertension, endocarditis, and so on, therefore the patient had a strong will to be treated after the baffle leak was confirmed. Because of the short time after the first thoracotomy (only 4 months) and the difficulty, severe trauma and high risks of second thoracotomy, such as post-operative adhesion, bleeding, respiratory failure, pulmonary infection, pulmonary atelectasis, pleural effusion, and so on, we finally reached an agreement with cardiac surgeons and decided to treat the baffle leak through percutaneous transcatheter closure, which has the advantages of lesser trauma and haemorrhage, slight pain, and rapid recovery. The patient was informed and willing to be treated by this method. After the interventional therapy, the patient’s symptom improved significantly, diastolic BP increased from 54 mmHg to 68 mmHg, which means improved coronary perfusion after the baffle leak closure, and PAP decreased due to shunt closure and probably improved cardiac function. To our knowledge, this is the first reported case of transcatheter closure for baffle leak after Takeuchi repair. There are still no available relevant articles at home and abroad we can learn from. Our initial experience is that: (i) make clear the relationship between the fistula and the ostium of coronary, evaluate the feasibility of percutaneous transcatheter closure, (ii) interventional procedure should be very soft to avoid the injury of coronary, (iii) avoid the occlusion of coronary after the implantation of occluder. But there are still some problems have to be addressed, for example, what kind of baffle leak can be treated by percutaneous transcatheter closure? What type of occluder can be used for baffle leak? What size of occluder should be recommended?

**Figure 1** Before the Takeuchi repair, computed tomography angiography shows that the left coronary artery arises from the main pulmonary artery.

**Figure 2** Four months after the Takeuchi repair, computed tomography angiography confirms a fistulous connection from the baffle into the main pulmonary artery, the diameter of the fistula is about 4 mm (A, red arrow); after the interventional therapy, the red arrow shows the device (B).
Conclusion

Percutaneous transcatheter closure is an alternative treatment option for the baffle leak after Takeuchi repair of ALCAPA, this case is just an initial exploration, more experience, and clinical data are still needed.

Supplementary material

Supplementary material is available at European Heart Journal – Case Reports online.

Consent: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: none declared.

References

1. Ginde S, Earing MG, Bartz PJ, Cava JR, Tweddell JS. Late complications after Takeuchi repair of anomalous left coronary artery from the pulmonary artery: case series and review of literature. Pediatr Cardiol 2012;33:1115–1123.
2. Peña E, Nguyen ET, Merchant N, Dennis C. ALCAPA syndrome: not just a pediatric disease. RadioGraphics 2009;29:553–565.
3. Cochrane AD, Coleman DM, Davis AM, Brizard CP, Wolfe R, Karl TR. Excellent long-term functional outcome after an operation for anomalous left coronary artery from the pulmonary artery. J Thorac Cardiovasc Surg 1999;117:332–342.
4. Schwartz ML, Jonas RA, Colan SD. Anomalous origin of left coronary artery from pulmonary artery: recovery of left ventricular function after dual coronary repair. J Am Coll Cardiol 1997;30:547–553.
5. Michielon G, Di Carlo D, Brancaccio G, Guccione P, Mazzera E, Toscano A, Di Donato RM. Anomalous coronary artery origin from the pulmonary artery: correlation between surgical timing and left ventricular function recovery. Ann Thorac Surg 2003;76:581–588.
6. Ojala T, Salminen J, Happonen J-M, Pihkala J, Jokinen E, Sairanen H. Excellent functional result in children after correction of anomalous origin of left coronary artery from the pulmonary artery—a population-based complete follow-up study. Interact Cardiovasc Thorac Surg 2010;10:70–75.
7. Yu-Juan Q, Pei-Jun L. Advancement of surgical therapy for anomalous left coronary artery from pulmonary artery. Chin J Thorac Cardiovasc Surg 2017;24:239–242.
8. Takeuchi S, Imamura H, Katsumoto K, Hayashi I, Katohgi T, Yozu R, Ohkura M, Inoue T. New surgical method for repair of anomalous left coronary artery from pulmonary artery. J Thorac Cardiovasc Surg 1979;78:7–11.
9. Dua R, Smith JA, Wilkinson JL, Menahem S, Karl TR, Goh TH, Mee RBB. Long-term follow-up after two coronary repair of anomalous left coronary artery from the pulmonary artery. J Cardiovasc Surg 1993;38:384–390.
10. Birk E, Stanler A, Katz J, Berant M, Dagan O, Matiau A, Erez E, Bleden LC, Vidne BA. Anomalous origin of the left coronary artery from the pulmonary artery: diagnosis and postoperative follow up. Isr Med Assoc J 2002;4:111–114.
11. Quanlin M, Jie X. Surgical treatment of anomalous origin of the left coronary artery from the pulmonary artery. Chin J Ethnopharmacol 2013;15:42–43.

Table 1  Summary of studies on baffle leak after Takeuchi repair of anomalous origin of the left coronary artery from the pulmonary artery

| Study         | Year | No. of patients | Baffle leak | Reoperation |
|---------------|------|-----------------|-------------|-------------|
| Schwartz et al. | 1997 | 21              | 11          | 3           |
| Cochrane et al. | 1998 | 12              | 2           | 0           |
| Birk et al.    | 2000 | 7               | 1           | 1           |
| Michielon et al. | 2003 | 12              | 1           | 1           |
| Ojala et al.   | 2009 | 4               | 1           | 0           |
| Ginde et al.   | 2012 | 9               | 3           | 1           |
| Quanlin and Jie | 2013 | 7               | 1           | 1           |
| Total (%)      |      | 72              | 20 (27.8)   | 7 (9.7)     |