

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

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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.

Introduction

Anomalous origin of the left coronary artery from the pulmonary artery (ALCAPA) is a rare but serious congenital coronary abnormality that if left untreated, results in high mortality.¹ There are two types of ALCAPA: the infant type and the adult type, each of which has different clinical manifestations and outcomes, which were described in detail by Peña *et al.*² Once ALCAPA is diagnosed, early surgical treatment is vital to correct the anomaly and prevent complications. The preferred surgical method is restoring a two-coronary-artery circulation system,^{1,2} and Takeuchi repair is an alternative treatment method for ALCAPA in certain situations.^{1,3} Late complications after Takeuchi repair of ALCAPA were reviewed by Ginde *et al.*, and it is

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reported that baffle leak is the most common complication.¹ 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 left anterior oblique (LAO) view and selective coronary angiography of the left main coronary artery 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 Q_p/Q_s 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.^{1,3-7} 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.^{1,8,9} Reported complications of Takeuchi repair mainly include baffle leak and supraventricular pulmonary stenosis, and the baffle leak is the most common complication (27%).^{1,4} 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.*,¹ 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

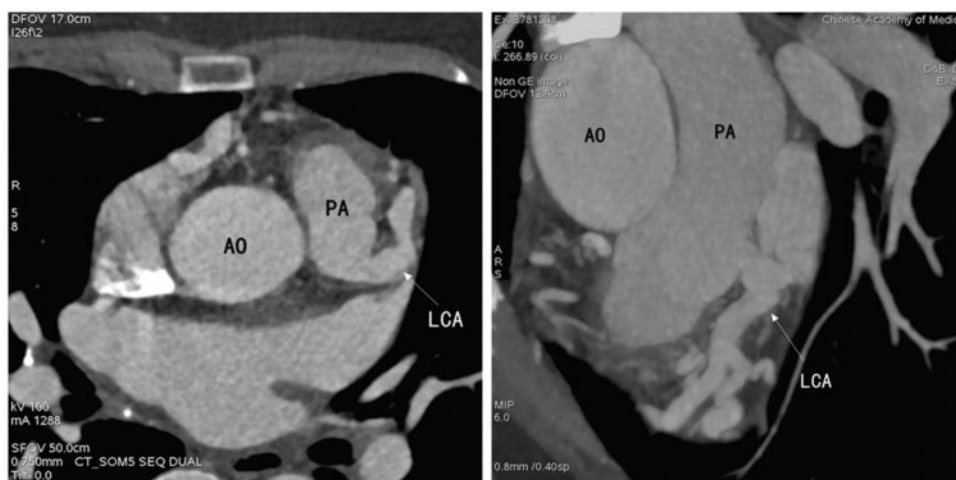


Figure 1 Before the Takeuchi repair, computed tomography angiography shows that the left coronary artery arises from the main pulmonary artery.



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).

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?

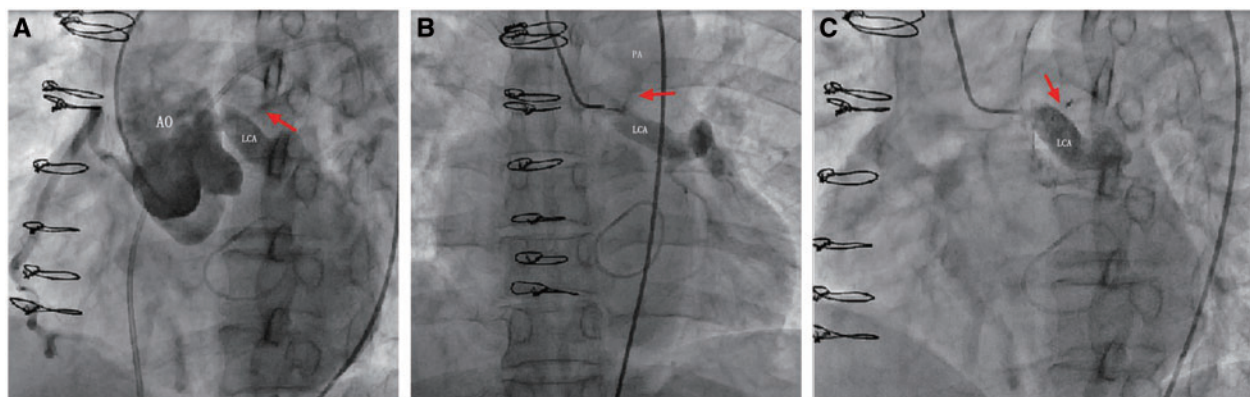


Figure 3 Percutaneous transcatheter closure: ascending aortic angiography in the left anterior oblique view (A) and selective coronary angiography of the left main coronary artery (B) demonstrates contrast leakage from the left coronary artery into the main pulmonary artery (red arrow); selective coronary angiography of the left main coronary artery in the left anterior oblique view (C) demonstrates no contrast leakage after implantation of the device (red arrow).

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. ⁴	1997	21	11	3
Cochrane et al. ³	1998	12	2	0
Birk et al. ¹⁰	2000	7	1	1
Michielon et al. ⁵	2003	12	1	1
Ojala et al. ⁶	2009	4	1	0
Ginde et al. ¹	2012	9	3	1
Quanlin and Jie ¹¹	2013	7	1	1
Total (%)		72	20 (27.8)	7 (9.7)

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.

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