Standardization of operations for coronary to pulmonary artery fistulas

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Abstract

Background: For coronary to pulmonary artery fistulas, operations are often indicated by the existence of myocardial ischemia or aneurysmal formations. Because lesions do not form in identical ways, a variety of surgical procedures are performed. We propose standardizing this operation for a safer and more reliable outcome.

Methods: We evaluated 8 cases of operations for coronary to pulmonary artery fistulas. A concrete standard method for this operation was sought from the case results.

Results: Standardization is summarized as: 1) Precise analysis of preoperative coronary angiography. 2) Thorough closure of fistulas and aneurysms. Cardiac arrest is preferred. 3) Perform as less pulmonary arteriotomy as possible. Use bicaval cannulation just in case of pulmonary arteriotomy. 4) During antegrade cardioplegic administration, oppress the fistulas to prevent stealing of solution. Use antegrade cardioplegia to confirm closure and hemostasis of fistulas and aneurysms. 5) Also use retrograde cardioplegia to ensure myocardial protection.

Conclusions: Because there are variations among lesions of coronary to pulmonary artery fistulas, we propose some practical methods that can be applied in common to take account of such anomalies.

Keywords: Coronary to pulmonary artery fistula, aneurysmal formation, standardization, pulmonary arteriotomy

Introduction

Most cases of coronary arteriovenous fistulas are congenital in origin, although acquired cases also exist [1,2]. Types draining into the pulmonary artery, which consist of 15% to 43% of surgical cases [1,3,4], are typical of these fistulas. Although most of these fistulas are tortuous vessels occasionally accompanied by aneurysms [5], the location of the connection to the coronary arteries, route, size, and amount of fistulas and the connection to the pulmonary artery are not identical. Thus, various surgical procedures are performed; for example, closure of fistulas under cardiac arrest, closure without cardiopulmonary bypass, closure of drainage to pulmonary artery from exterior side, and closure under pulmonary arteriotomy. For a safer and more reliable outcome, we propose standardizing the operative method.

Methods

A total of 8 operations for coronary to pulmonary artery fistulas were performed. We evaluated patient profiles, operative methods and results.

Patient profiles are summarized in (Table 1). 8 patients with the age and gender indicated received surgery. 4 patients were asymptomatic, 3 patients were angina and 1 patient was dyspnea on exertion. 3 of the asymptomatic patients showed ischemia in electrocardiography, and 1 patient was referred due to cardiac murmur. Upon performing coronary angiography, 7 patients were diagnosed as having aneurysmal formations, including 1 having 2 aneurysms. The patient that did not have an aneurysm was operated upon for angina and ischemia. The patient with dyspnea on exertion had no ischemia, but received surgery for a 2-cm aneurysm. The patient referred with cardiac murmur also had no ischemia, but received surgery for a 2-cm aneurysm.

(Table 2) summarizes operative methods and results. Concomitant coronary artery bypass grafting (CABG) was performed in 4 patients, of whom 2 underwent 2 bypasses and another 2 underwent 1 bypass each. All operations were performed under cardiopulmonary bypass. Bicaval cannulation was used in 6 patients just in case pulmonary arteriotomy was needed to close the fistulas. The other 2 patients had the entrance of the fistulas to the pulmonary artery at the anterior side, and closure under pulmonary arteriotomy. For a safer and more reliable outcome, we propose standardizing the operative method.

Conclusions

Because there are variations among lesions of coronary to pulmonary artery fistulas, we propose some practical methods that can be applied in common to take account of such anomalies.

Keywords: Coronary to pulmonary artery fistula, aneurysmal formation, standardization, pulmonary arteriotomy

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were studied from many directions. *(Figure 2)* shows intraoperative pictures of the same patient. Careful observation of the fistulas and the aneurysms together with findings of coronary angiography in mind direct us to the vessels to be closed. The fistulas are closed thoroughly with pledgetted stitches and sometimes with clips. It is important to identify as many fistulas as possible and to close all fistulas however small. The aneurysms are closed by opening them, identifying their inflows and outflows, and suturing or ligating those inflows and outflows. In order to be certain of their closure, aneurysmectomy is performed where possible. Some cases of small aneurysms were crushed with sutures. When aneurysms were present near the pulmonary artery, a side clamp was applied to the pulmonary artery to facilitate closure.

From experience with these 8 cases, a concrete standard surgical method for coronary to pulmonary artery fistulas was sought. Based on the excellent results obtained, we have confidence in our methods. Standard planning and principles that can be applied in common are proposed in the results section below.

### Results

First, a decision tree was made for the formation of aneurysms and the need for CABG *(Figure 3).* Because aneurysmal formations often coexist with these fistulas, it is inevitable for these aneurysms to be located in the main stem.

The results of standardization are as follows:

1. Precise analysis of preoperative coronary angiography. Coronary computed tomographic angiography may be substituted, if analyzed precisely. These analyses include careful assessment of the inflow and outflow of fistulas and their relationships with coronary and pulmonary arteries from many directions. They also include the inflow and outflow of aneurysmal formations and their relationships with fistulas.

2. Thorough closure of fistulas and aneurysms. This procedure to close the fistulas takes time. Cardiac arrest is preferred especially when there are aneurysms *(Figure 3).* When there are no aneurysms and CABG is needed, cardiopulmonary bypass can be omitted depending on the case *(Figure 3).* Cases without aneurysmal formation and without the need for CABG are the best candidates to be closed without cardiopulmonary bypass *(Figure 3).*

3. Perform as less pulmonary arteriotomy as possible. Most of the entries to the pulmonary artery are closed by occluding fistulas seen from the exterior side. If there are difficulties such as the presence of an aneurysm nearby, use side clamps to facilitate closure. Use bicaval cannulation just in case of pulmonary arteriotomy.

4. During antegrade cardioplegic administration, oppress the fistulas digitally or clamp them with instruments to prevent stealing of solution. Use antegrade cardioplegia to confirm closure and hemostasis of fistulas and aneurysms. If fistulas remain, they will dilate during administration.

5. Also use retrograde cardioplegia to ensure myocardial protection. It is not only a matter of stealing of the antegrade cardioplegia. Combining retrograde cardioplegia saves time because there is no need to interrupt the operative procedure. Retrograde cardioplegia does not usually flow into the fistulas and interrupt surgery.

### Discussion

Operations for coronary to pulmonary artery fistulas are indicated in the presence of myocardial ischemia, aneurysmal formations or heart failure due to shunt flow. They are also performed in asymptomatic patients when future symptoms are anticipated. Generally, surgery for a lesion is safe to perform, which means the procedure has to be performed without complications, especially in asymptomatic patients. Besides, the results must leave as few fistulas as possible to

| Table 1. Patient profiles. |
|---------------------------|
| **Patient profiles** |
| Number | 8 |
| Age | 55.4 ± 11.9 years |
| Gender | 6 males; 2 females |
| Symptoms |
| asymptomatic | 4 |
| angina | 3 |
| dyspnea on exertion | 1 |
| Surgical Indications |
| ischemia + aneurysm | 5 |
| aneurysm | 2 |
| ischemia | 1 |
| Aneurysmal Size |
| 1 – 2 cm | 6 |
| 3 – 4 cm | 2 (in one patient ) |

Age is expressed as mean ± standard deviation.

| Table 2. Operative methods and results. |
|-------------------------------|
| **Operative methods** | **Results** |
| Concomitant CABG |
| two bypasses | 2 |
| one bypass | 2 |
| none | 4 |
| Use of Cardiopulmonary Bypass |
| Bicaval Cannulation | 8 |
| Pulmonary Arteriotomy | None |
| Cardiac Arrest Time |
| with CABG | 104.3 ± 13.8 minutes |
| without CABG | 50.0 ± 19.1 minutes |
| Residual Fistulas | None |
| Morbidity and Hospital Deaths | None |

CABG: Coronary Artery Bypass Grafting
Cardiac Arrest Time is expressed as mean ± standard deviation.
achieve the goal of the operation. Because there are variations among lesions, we considered that some standardization of the procedure would make the results sufficiently reliable. Thus, 8 cases were analyzed.

The first point is a precise analysis of preoperative coronary angiography. As stated by Lazar et al. [5], especially when there is a large aneurysm complicated with fistulas, preoperative imaging yields the safest course of resection. We must be aware of the fact that despite multiple imaging methods being available for use, defining the aneurysm’s exact vasculature can be challenging [5]. Because coronary angiography and coronary computed tomographic angiography are the best and only methods for deciding preoperative strategy, these must be studied precisely. The inflow and outflow of fistulas and their relationships with coronary and pulmonary arteries, as well as the inflow and outflow of aneurysmal formations and their relationships with fistulas, must be studied from many directions.

The apparent consensus is that surgical resection under sternotomy and cardiopulmonary bypass is the best long-term solution, especially in cases with aneurysms [5]. A standard treatment protocol has not been established and other methods such as coil embolization are not preferred especially in symptomatic patients [5]. This means that surgical closure of

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**Figure 1.** (A) Preoperative coronary angiography. The arrow indicates a large tortuous coronary to pulmonary artery fistula originating from the left anterior descending artery (LAD). There was a 90% stenosis in the proximal LAD. Arrowheads indicate 2 aneurysms. (B) Postoperative coronary angiography. Coronary to pulmonary fistulas are closed, and the distal LAD not observed due to a patent bypass to the LAD.

**Figure 2.** Intraoperative pictures of the same patient as in Figure 1. The bottom of the pictures show the direction of the patient’s head. (A) Before closure of fistulas and aneurysms. The arrow indicates the tortuous coronary to pulmonary artery fistula, and the arrowheads indicate aneurysms. (B) After closing the fistulas and aneurysms. Thorough closure was performed using pledgetted sutures. The left internal thoracic artery was bypassed to the left anterior descending artery.
fistulas must be performed safely, with reliable results, which can be performed under an arrested heart. Cardiopulmonary arrest and use of antegrade cardioplegia are essential for confirming closure of fistulas and hemostasis, as mentioned.

We recommend oppressing the fistulas to prevent stealing of solution during antegrade cardioplegic administration, as stated by some authors [1, 6, 7]. For the same reason, Maeda et al., plan to close as many fistulas as possible before initiating cardiopulmonary bypass in their operations [8]. We do not deny there are simple cases in which fistulas can be closed under a beating heart, but such cases are seldom indicated for surgery because they can be closed by coil embolization. Many cases are complicated with aneurysms and the need for concomitant CABG.

Others state that antegrade cardioplegia should be used to test the security of a closure [4]. We add another reason, which is to confirm hemostasis of fistulas and aneurysms. Others also agree with our recommendation to use retrograde cardioplegia [6]. This ensures myocardial protection in the presence of coronary to pulmonary artery fistulas where stealing may occur during antegrade administration.

Some authors open the pulmonary artery to close the entry points of the fistulas with a patch of autologous pericardium [9]. Although this method may be effective to ensure that all multiple openings are addressed, we consider that all small openings cannot be identified from inside and the same effect can be obtained by closing fistulas from the exterior side. Although we found literature that advocated closing these fistulas from within the pulmonary artery cavity [10], we consider that the procedure for opening this artery is seldom necessary. From our experience, we can use side clamps to close entry points to the pulmonary artery, if necessary. There are studies that insist on closing fistulas with an external approach [2], which is similar to our method.

Conclusions
Coronary to pulmonary artery fistulas have diverse lesions. They are often combined with aneurysmal formations and surgical indication does not always include symptoms. Safe and reliable results are demanded, so we propose a standard procedure including cardiac arrest and closure from the exterior side of the pulmonary artery.

Competing interests
The authors declare that they have no competing interests.

Authors’ contributions
ST, AM & TN contributed to the pre-operative planning and operation of the cases. All authors contributed to following up the cases. All authors read and approved the final manuscript.

Acknowledgement
Authors would like to acknowledge the perfusionists Mr. Kazutoshi Nakao, Mr. Takeshi Sato and Ms. Megumi Imai for taking and providing the intraoperative pictures.

Publication history
EJC: William Clifford Roberts, Baylor University Medical Center, USA. Received: 06-May-2013 Revised: 02-Jun-2013 Re-Revised: 09-Jul-2013 Accepted: 16-Jul-2013 Published: 22-Jul-2013

References
1. Mangukia CV. Coronary artery fistula. Ann Thorac Surg. 2012; 93:2084-92. | Article | PubMed
2. Onorati F, Mastroroberto P, Bilotta M, Cristodoro L, Esposito A, Pezzo F and Renzulli A. Surgical treatment of coronary-to-pulmonary fistula: how and when? Heart Vessels: 2006; 21:321-4. | Article | PubMed
3. Huang YK, Lei MH, Lu MS, Tseng CN, Chang JP and Chu JJ. Bilateral coronary-to-pulmonary artery fistulas. Ann Thorac Surg. 2006; 82:1886-8. | Article | PubMed
4. Kirklin JW, Barratt-Boyes BG, Blackstone EH, Jonas RA and Kouchoukos NT. Congenital Anomalies of the Coronary Arteries. In Kirklin JW and Barratt-Boyes BG (Eds.), Cardiac Surgery. 1993; 1167-93. | Book
5. Lazar JF, Compton M, Li F and Knight P. Excising a giant: report of a 7-cm coronary artery aneurysm. Tex Heart Inst J. 2013; 40:173-5. | PubMed
6. Hajj-Chahine J, Belmonte R, Lefant PY and Tomasi J. eComment: Cardiopulmonary in coronary artery fistula to coronary sinus. Int J Cardiovasc Thorac Surg. 2011; 13:675. | Article | PubMed
7. Hajj-Chahine J, Haddad F, El-Rassi I and Jebraa V. Surgical management of a circumflex aneurysm with fistula to the coronary sinus. Eur J Cardiothorac Surg. 2009; 35:1086-8. | Article | PubMed
8. Maeda K, Yoshikawa Y, Miyagawa S, Nishi H, Fukushima S, Ueno T, Toda K, Kuratani T and Sawa Y. Surgical Treatment of Coronary Arteriovenous Fistulas and Aortic Valve Insufficiency. J Card Surg. 2013. | Article | PubMed
9. Dimitrakakis G, Von Oppell U, Luckraz H and Groves P. Surgical repair of triple coronary-pulmonary artery fistulae with associated atrial septal defect and aortic valve regurgitation. Interact Cardiovasc Thorac Surg. 2008; 7:933-4. | Article | PubMed
10. Fernandes ED, Kadivar H, Hallman GL, Reul GJ, Ott DA and Cooley DA. Congenital malformations of the coronary arteries: the Texas Heart Institute experience. Ann Thorac Surg. 1992; 54:732-40. | Article | PubMed

Taguchi S, Mori A, Suzuki R, Ishida O and Niibori T. Standardization of operations for coronary to pulmonary artery fistulas. Cardio Vasc Syst. 2013; 1:7. http://dx.doi.org/10.7243/2052-4358-1-7