Tushar C. Barot, MD Angelo LaPietra, MD Orlando Santana, MD Nirat Beohar, MD Joseph Lamelas, MD

Key words: Arteriovenous malformations/physiopathology; coronary artery bypass/ adverse effects; fistula/etiology; internal mammary-coronary artery anastomosis/ adverse effects; mammary arteries/surgery; myocardial ischemia/etiology; pulmonary artery/surgery; vascular fistula/complications/etiology/surgery

From: Division of Cardiac Surgery (Drs. Barot, Lamelas, and LaPietra) and Columbia University Division of Cardiology (Drs. Beohar and Santana), Mount Sinai Heart Institute, Miami Beach, Florida 33140

#### Address for reprints:

Orlando Santana, MD, Columbia University Division of Cardiology, Mount Sinai Heart Institute, 4300 Alton Rd., Miami Beach, FL 33140

#### E-mail:

osantana@msmc.com

© 2014 by the Texas Heart® Institute, Houston

# Multiple Left Internal Mammary Artery-to-Pulmonary Artery Fistulae

15 Years after Coronary Artery Bypass Grafting

Left internal mammary artery (LIMA)-to-pulmonary artery fistulae rarely develop after coronary artery bypass grafting. Fewer than 30 cases of these fistulae have been reported since 1947. Nevertheless, this entity should be considered as a cause of recurrent angina after bypass surgery, in the absence of other causes. We present the case of a 67-year-old man with cardiac symptoms in whom multiple LIMA-to-pulmonary artery fistulae were found, 15 years after he had undergone coronary artery bypass grafting. The diagnosis was confirmed by means of coronary angiography with selective catheterization of the LIMA and by computed tomographic angiography of the heart. The patient underwent reoperative 2-vessel coronary artery bypass grafting and ligation of multiple fistulae; 16 months postoperatively, he was asymptomatic and doing well. In addition to reporting this case, we discuss relevant diagnostic and treatment considerations. **(Tex Heart Inst J 2014;41(1):94-6)** 

s a bypass-graft conduit to the left anterior descending coronary artery (LAD), the left internal mammary artery (LIMA) is the vessel of choice because of its proven longevity and long-term patency. The formation of a LIMA-to-pulmonary artery (PA) fistula after coronary artery bypass grafting (CABG) is a rare complication: fewer than 30 reports have appeared in the medical literature.<sup>1</sup> We report a case of multiple LIMA-to-PA fistulae that we found 15 years after a patient had undergone CABG.

## **Case Report**

A 67-year-old white man presented at our institute in 1997 with multivessel coronary artery disease. At that time, he underwent 5-vessel CABG, with the following grafts: LIMA to LAD, saphenous vein (SV) to the first diagonal artery, sequential SV to the first and 3rd obtuse marginal branches, and SV to the distal right coronary artery (RCA). The patient's recovery was uneventful. In 2006, he underwent percutaneous stenting of the native RCA. In 2012, he reported chest tightness, exertional dyspnea, and general weakness. He underwent a full cardiac examination, including coronary angiography. The chief findings were an occluded SV graft to the obtuse marginal artery and an 80% stenosis of the native RCA, distal to the previously placed stent. The LIMA-to-LAD graft was patent. However, there were multiple large fistulae from the LIMA to the left PA, with brisk contrast washout into the PA upon selective LIMA injection during cardiac catheterization (Figs. 1 and 2). A computed tomographic angiogram of the heart revealed a markedly tortuous LIMA with tangled vessels in the prevascular space of the anterior mediastinum. Multiple scattered, tortuous subsegmental branches of the anterior left PA throughout the anterior aspect of the left upper lobe appeared to connect to the anterior mediastinal vessels, confirming the diagnosis of multiple LIMA-to-left PA fistulae (Fig. 3).

The numerous small and large fistulous communications between the LIMA and the left PA were not suitable for percutaneous intervention, so the patient underwent 2-vessel CABG: a right internal mammary artery graft to the LAD, and a SV graft to the RCA. During surgery, we saw multiple fistulae that extended from the LIMA to the left PA. Smaller branches were clipped; larger branches were ligated and transected. The patient tolerated the procedure well, his postoperative course was uneventful, and he was discharged from the hospital on postoperative day 7. Sixteen months postoperatively, he was asymptomatic and doing well.

## Discussion

Fistulae from the LIMA to the PA are a rare clinical phenomenon. The first case of such fistulae was described



**Fig. 1** Coronary angiogram (left anterior oblique view) shows the left internal mammary artery (LIMA) graft with 3 large fistulae and multiple smaller fistulae to the pulmonary artery.

by Burchell and Clagett<sup>2</sup> in 1947; since then, fewer than 30 other relevant case reports have been published. In 595 post-CABG patients, only 0.67% developed a similar fistula.<sup>1</sup> The average developmental time for fistulae is approximately 5 years (range, 2 mo–13 yr).<sup>1,3,4</sup>

A LIMA-to-PA fistula can be congenital or acquired. The acquired form can be secondary to trauma, neoplasm, or inflammatory disease.<sup>5-10</sup> The causal mechanism is not fully understood. Predisposing factors for the development of fistulae include injury to the pleura and lung parenchyma, incomplete ligation of the intercostal branches of the LIMA, the use of electrocautery instead of ligation of side branches, infection, and inflammation leading to neovascularization.<sup>11-18</sup> Some authors have suggested meticulous dissection of the LIMA to avoid injury to the pleura and lung parenchyma, clipping or ligating the arterial branches instead of using electrocautery to prevent recanalization, and covering the LIMA with a pericardial flap to prevent fistula formation.<sup>13,14,19,20</sup>

These fistulae can result in substantial shunting of blood from the LIMA to the PA, causing myocardial ischemia.<sup>1</sup> The most often reported presenting symptom is recurrent angina; however, the spectrum can encompass dyspnea, congestive heart failure, and end-arteritis in the presence of a continuous murmur.<sup>12,16,21-23</sup> The most severe complication, coronary-to-pulmonary steal, can be fatal.<sup>19,24</sup> Every patient who presents with any of the above symptoms after CABG should undergo a complete cardiac examination. The diagnostic test of choice for LIMA-to-PA fistula is cardiac catheter-ization with selective angiography.<sup>13,21,25</sup>



**Fig. 2** Coronary angiogram (right anterior oblique view) shows the left internal mammary artery (LIMA) graft to the left anterior descending coronary artery, and multiple fistulae to the pulmonary artery (PA).



**Fig. 3** Computed tomographic angiogram (3-dimensional reconstruction) shows multiple fistulae from the left internal mammary artery (LIMA) graft to subsegmental branches of the anterior left upper lobe of the pulmonary artery (PA), draining into the PA.

Management options for LIMA-to-PA fistulae include conservative medical therapy, coil embolization, percutaneous angioplasty with stenting, and surgical ligation.<sup>20-23,25,26</sup> No one treatment yields clear benefits; the management approach depends on physician and institutional experience. Some authors recommend starting with conservative management and progressing to more invasive treatment options,<sup>23</sup> whereas others advocate a conservative approach over invasiveness except when the fistula causes substantial symptoms or grows in size.<sup>15</sup> We think that the choice of treatment should be based on the patient's symptoms and the physician's experience.

### References

- Madu EC, Hanumanthu SK, Kim C, Prudoff A. Recurrent ischemia resulting from left internal mammary artery-to-pulmonary artery fistula. Angiology 2001;52(3):185-8.
- Burchell HB, Clagett OT. The clinical syndrome associated with pulmonary arteriovenous fistulas, including a case report of a surgical cure. Am Heart J 1947;34(2):151-62.
- Hakeem A, Bhatti S, Williams EM, Biring T, Kosolcharoen P, Su Min C. Coronary steal due to bilateral internal mammary artery--pulmonary artery fistulas: a rare cause of chest pain after coronary artery bypass grafting. Angiology 2008;59(2): 244-7.
- Maiello L, Franciosi G, Presbitero P, Gallotti R. Left internal mammary artery to pulmonary artery fistula after minimally invasive coronary bypass. Ann Thorac Surg 2002;73(1):317.
- Senno A, Schweitzer P, Merrill C, Clauss R. Arteriovenous fistulas of the internal mammary artery. Review of the literature. J Cardiovasc Surg (Torino) 1975;16(3):296-301.
- Ruberti U, Odero A, Arpesani A, Giorgetti PL, Cugnasca M, Rampoldi V, Anguissola GB. Internal mammary artery to pulmonary artery fistula. J Cardiovasc Surg (Torino) 1986; 27(6):734-6.
- Hearne SF, Burbank MK. Internal mammary artery-to-pulmonary artery fistulas. Case report and review of the literature. Circulation 1980;62(5):1131-5.
- Brundage BH, Gomez AC, Cheitlin MD, Gmelich JT. Systemic artery to pulmonary vessel fistulas: report of two cases and a review of the literature. Chest 1972;62(1):19-23.
- 9. Cohen EM, Loew DE, Messer JV. Internal mammary arteriovenous malformation with communication to the pulmonary vessels. Am J Cardiol 1975;35(1):103-6.
- Dunn RP, Wexler L. Systemic-to-pulmonary fistula in intrapulmonary Hodgkin's disease. Chest 1974;66(5):590-4.
- 11. Groh WJ, Hovaguimian H, Morton MJ. Bilateral internal mammary-to-pulmonary artery fistulas after a coronary operation. Ann Thorac Surg 1994;57(6):1642-3.
- Imawaki S, Arioka I, Nakai M, Tsuruno Y, Takama T, Maeta H, Inagawa T. Development of a fistula between an internal mammary artery graft and the pulmonary vasculature following coronary artery bypass grafting: report of a case. Surg Today 1995;25(5):461-4.
- Blanche C, Eigler N, Bairey CN. Internal mammary artery to lung parenchyma fistula after aortocoronary bypass grafting. Ann Thorac Surg 1991;52(1):141-2.
- Ferreira AC, Marchena E, Liester M, Sangosanya AO. Internal mammary to pulmonary artery fistula presenting as early recurrent angina after coronary bypass. Arq Bras Cardiol 2002;79(2):181-2.

- Guray U, Guray Y, Ozbakir C, Yilmaz MB, Sasmaz H, Korkmaz S. Fistulous connection between internal mammary graft and pulmonary vasculature after coronary artery bypass grafting: a rare cause of continuous murmur. Int J Cardiol 2004; 96(3):489-92.
- Almeida Junior GL, Jorge JK, Neno AC, Nogueira FB, Hellmuth B, Lins RH, et al. Left internal thoracic artery to left pulmonary artery fistula after coronary artery bypass graft surgery. A rare cause of myocardial ischemia [in Portuguese]. Arq Bras Cardiol 2005;85(5):337-9.
- 17. Nellens P, Stevens C, Verstraeten J, Heyndrickx GR. Internal mammary to pulmonary artery fistula associated with healed tuberculosis. Acta Cardiol 1980;35(1):55-61.
- Wood MK. Internal mammary artery to lung parenchyma fistula. Ann Thorac Surg 1992;54(3):603.
- Garrean S, Tshibaka C, Hanhan Z, Geha AS, Massad MG. Coronary-pulmonary steal caused by internal thoracic arterypulmonary artery fistula after coronary artery bypass operations. J Thorac Cardiovasc Surg 2005;130(2):569-71.
- Nielson JL, Kang PS. Endovascular treatment of a coronary artery bypass graft to pulmonary artery fistula with coil embolization. Cardiovasc Intervent Radiol 2006;29(2):302-5.
- Johnson JA, Schmaltz R, Landreneau RJ, Wright WP, Curtis JJ, Walls JT, Nawarawong W. Internal mammary artery graft to pulmonary vasculature fistula: a cause of recurrent angina. Ann Thorac Surg 1990;50(2):297-8.
- 22. Abbott JD, Brennan JJ, Remetz MS. Treatment of a left internal mammary artery to pulmonary artery fistula with polytetrafluoroethylene covered stents. Cardiovasc Intervent Radiol 2004;27(1):74-6.
- Peter AA, Ferreira AC, Zelnick K, Sangosanya A, Chirinos J, de Marchena E. Internal mammary artery to pulmonary vasculature fistula--case series. Int J Cardiol 2006;108(1):135-8.
- Najm HK, Gill IS, FitzGibbon GM, Keon WJ. Coronary-pulmonary steal syndrome. Ann Thorac Surg 1996;62(1):264-5.
- 25. Cay S. The left internal mammary artery to pulmonary vasculature fistula causing significant ischemia: which type of therapy is the best? Int J Cardiol 2010;144(3):453-4.
- Bijulal S, Namboodiri N, Nair K, Ajitkumar VK. Native vessel angioplasty as treatment strategy for left internal mammary artery to pulmonary vasculature fistula producing coronary steal phenomenon. Int J Cardiol 2009;133(1):e25-7.