



Pulmonary artery interventions after the arterial switch operation: Unique and significant risks

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Abstract

Background: In the modern era, results of the arterial switch operation (ASO) for transposition of the great arteries are excellent. However, because of the LeCompte maneuver, there may be a propensity for development of pulmonary artery stenosis. We encountered atypical complications of pulmonary artery stenting in patients after the ASO, including aorto-pulmonary fistula and coronary compression.

Methods: We performed a 10-year retrospective review of catheterizations performed in patients after ASO in our institution with a focus on adverse events.

Results: Diagnostic and interventional catheterizations were performed in 47 patients. In 29 patients, 37 interventional procedures performed, which included pulmonary artery angioplasty and/or stenting. In this group, there were five major adverse events (14%), including three aorto-pulmonary fistulae and one coronary artery compression among patients having stent implantation or stent redilation. In addition, there were 6/37 (16%) intended stent procedures, which were aborted because there appeared to be high-risk of significant adverse events.

Conclusions: This review suggests that percutaneous intervention on pulmonary artery stenosis after ASO has high-risk and should be undertaken advisedly. Prior thorough evaluation of coronary arteries is mandatory as coronary reimplantation sites may be adjacent to sites of pulmonary artery stenosis. Furthermore, if pulmonary artery stent implantation or stent redilation is contemplated, the risk of stent fracture and possible AP fistula should be recognized. Primary use of reinforced covered stents should be considered.

KEYWORDS

aorto-pulmonary fistula, arterial switch operation, pulmonary artery, stents, transposition of great vessels

1 | INTRODUCTION

Dextro-transposition of the great arteries (d-TGA) accounts for 5%-7% of congenital heart defects.¹ The arterial switch operation typically with the LeCompte maneuver (ASO) is the treatment of choice. This maneuver may involve "stretching" of the pulmonary artery branches while moving the pulmonary artery bifurcation

anterior to the proximal neoascending aorta. The anastomosis to the neomain pulmonary artery may be under "tension" despite the use of patch arterioplasty; pulmonary artery branches are splayed around the ascending aorta and may be compressed. The neopulmonary artery or branch pulmonary arteries may be in close proximity to sites where the coronary artery buttons are implanted into the neoascending aorta. These mechanical and anatomic features of the

LeCompte maneuver may predispose patients to develop neoinfarction or branch pulmonary artery stenosis, and in addition may create unique susceptibility to adverse consequences of stent procedures.

Although early mortality and morbidity are low,² and midterm results are excellent, many patients who undergo the ASO eventually require reintervention.³ Previous single center studies have reported incidences of pulmonary artery stenosis requiring intervention ranging from 3.8% to 28%.⁴⁻⁶ Pulmonary artery angioplasty and/or stenting have been the most common procedures performed in these patients.⁴⁻⁹

We encountered aorto-pulmonary (AP) fistulae and coronary artery compression during pulmonary artery stent procedures in this population. Therefore, we reviewed catheterization procedures performed in our institution after ASO. In an effort to determine whether these complications were unusual, we focused the review on adverse events and risk situations. Our aims were to identify types and frequency of interventions, complications, high-risk situations, and to determine likely associated risk factors.

2 | METHODS

We performed a retrospective review of patients at Rady Children's Hospital with d-TGA or double outlet right ventricle (DORV) post ASO who had cardiac catheterization over a 10-year period (January 2006 to December 2016). This study was approved by our Institutional Review Board.

The medical records of the patients were reviewed. Data abstracted included the age, gender, original diagnosis, surgical history, time to catheterization, indications for catheterizations, types and timing of interventions performed. Angiograms and other imaging were reviewed. Adverse events, high-risk situations, risk factors, event presentation and management, as well as patient outcomes were determined.

3 | RESULTS

During the study period, our surgeons performed an average of 15 neonatal ASO annually. We performed diagnostic and interventional catheterizations in 47 patients who had prior ASO. Indications for catheterization included early post-ASO persistent pleural effusions in three patients, coarctation of the aorta in six patients, and assessment of coronary arteries and hemodynamics in nine patients.

In 29 patients, we performed 37 procedures because of pulmonary artery stenosis. Among these patients, the primary diagnosis was d-TGA with intact IVS (19), d-TGA with ventricular septal defect (VSD) (5), d-TGA with DORV (4), and d-TGA with other lesions (1). Twenty-seven of the patients had neonatal arterial switch operations, and two had staged repairs. Twenty-five patients had their ASO at our institution and four at other institutions. All patients underwent an ASO with a LeCompte maneuver and five had patch pulmonary arterioplasty. Some patients underwent additional surgery

at various ages including patch pulmonary arterioplasty ($n = 10$), pulmonary valve replacement ($n = 4$).

Coronary anatomy in these patients was typical for transposition in 23. There was single coronary origin in two, Circumflex from the RCA in three, and one had intramural course of the LCA. High re-implantation of the RCA and or LCA was reported in three patients. Revision of the LCA anastomosis was performed a few months after the ASO in one patient.

At the time of catheterization, patient median age was 11.3 years. The median time between ASO and a catheterization procedure was 10.7 years. Twenty-three patients were male. Indications for catheterization were branch pulmonary artery stenosis ($n = 25$) and/or supra-valvar and valvar pulmonary stenosis ($n = 15$). One of these patients with supra-valvar stenosis also had a highly incompetent neopulmonary valve. Indications for catheterization were determined by echocardiogram. These included evidence of high-pressure RV determined by tricuspid regurgitation velocity or outflow gradient, RV enlargement and dysfunction, narrowing of the MPA or pulmonary branches by 2D assessment, and/or Doppler velocities suggesting greater than 30 mm Hg gradients in the MPA or pulmonary artery branches. At catheterization indications for interventions included RV pressure greater than half systemic, significant angiographic narrowing, and/or pressure gradients greater than 30 mm Hg.

In 37 catheterizations, the patients underwent angioplasty ($n = 16$) or stenting ($n = 21$) of pulmonary artery branches, angioplasty ($n = 16$) of main pulmonary artery, and/or percutaneous pulmonary valve implantation ($n = 1$). Fifteen (70%) of the 21 stented vessels involved various Palmaz stents (Cordis, Milpitas, California), 4 vessels involved DoubleStrut 16 mm stents (Covidien, ev3, Plymouth, Minnesota), and two stented vessels were unidentified from the medical record. Acute outcomes of catheterizations in these patients included reduction of RV systolic pressure from mean of 59.8 mm Hg (SD 14.7) to 47.5 mm Hg (SD 10.3) and angiographic improvement in minimum vessel diameter. During the study period 8 of the 29 patients required a second catheterization for intervention on the pulmonary arteries.

There were no deaths, urgent surgeries, or permanent injuries related to the catheterizations. Major adverse events with stent fractures in branch pulmonary arteries, noted at catheterization in 4 patients. Three of these patients had complicating AP fistulae develop at the sites of the stent fractures. Other major adverse events included left main coronary artery (LMCA) compression by a left pulmonary artery (LPA) stent in one case.

In addition, there were six high-risk situations in four additional patients who did not have major adverse events. Four patients had six episodes of threatened LMCA compression due to close proximity to the LPA (two patients, four episodes) or supra-valvar area (two patients, two episodes) during intended stenting.

3.1 | Aorto-pulmonary fistulae

In the three patients with AP fistulae, two were from the LPA (Figure 1) and one was from the right pulmonary artery (RPA) (Figure 2). All cases were identified to have stent fractures at the

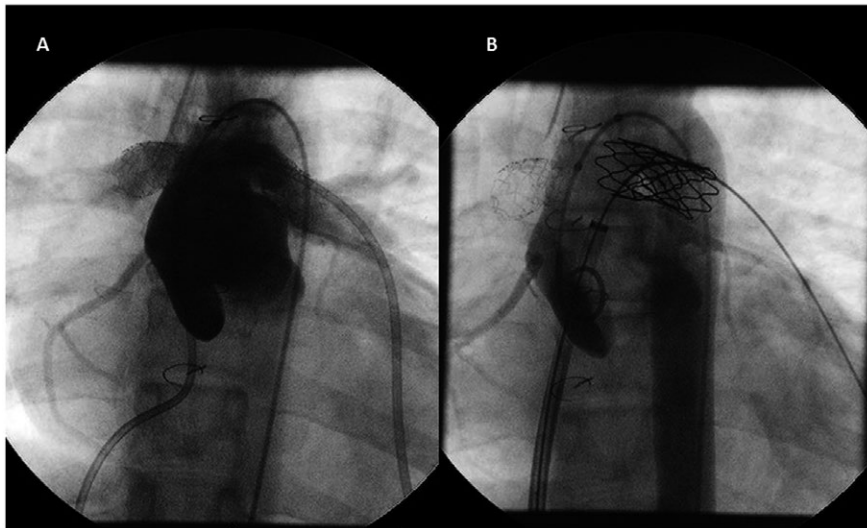


FIGURE 1 Twelve-year-old female status post arterial switch operation as an infant and subsequent bilateral pulmonary artery branch stenosis requiring stenting and dilations, who developed new continuous murmur. A, Aortogram reveals an AP fistula from the ascending aorta to the left pulmonary artery at a stent fracture site. B, The fistula is closed with a covered stent.

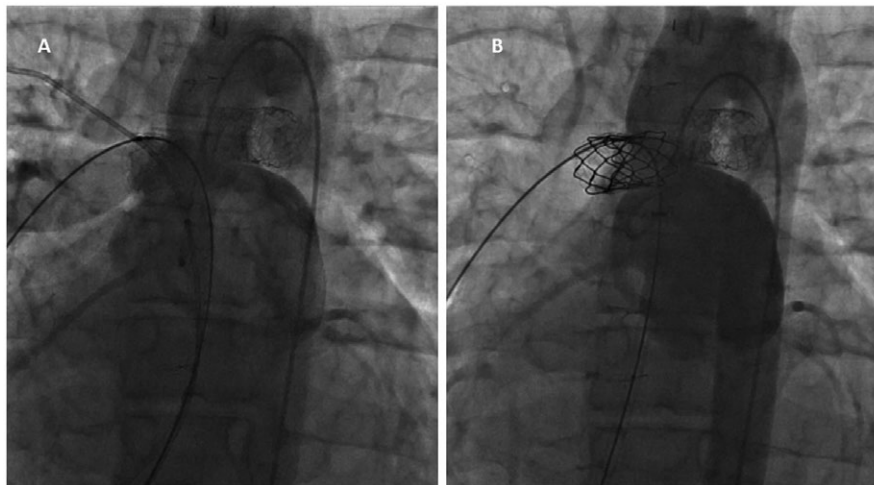


FIGURE 2 Seventeen-year-old male status post arterial switch operation as an infant and subsequent bilateral pulmonary artery branch stenosis requiring surgical revision and bilateral pulmonary stents, who was noted to have an aorta to right pulmonary artery fistula following balloon dilation and stent fracture by angiography. A, Aortogram reveals an AP fistula from the ascending aorta to the right pulmonary artery at a stent fracture site. B, The fistula is closed with a covered stent.

fistula site. Stent fractures resulted from various stent manufacturers. Time after ASO in these cases was 10-17 years. All AP fistulae were treated by implantation of covered stents in the involved pulmonary artery branches. After covered stent implantations, there were no residual shunts (Figures 1 and 2; Table 1). Follow-up of these patients 2-5 years post implant of covered stents has suggested no residual fistulae by examination or echocardiography.

Patient 1: A 13-year-old underwent ASO as a neonate with surgical patch arterioplasty of his branch pulmonary arteries and LPA stent implant at age nine. A year later at an outside institution, a stent was placed in the RPA and the previously placed LPA stent was dilated to 14 mm. This dilation caused two major complications. The first was coronary compression requiring emergent coronary

stenting (see description in below section). Secondly, a LPA stent fracture was noted along with a large AP fistula resulting in heart failure. The patient was referred to our institution. We closed the fistula with a CP covered stent (NuMED, Hopkinton, New York).

Patient 2: A 17-year-old underwent ASO as a neonate. The patient subsequently had stents placed in bilateral proximal branch arteries with DoubleStrut 16 mm stents (Covidien, ev) at 2 years of age. The pulmonary arteries were restented at age 14 with a Palmaz Genesis 1910B stent in the RPA and a Palmaz Genesis 2910B stent (Cordis) in the LPA. The stents were dilated 3 years later with an Atlas 16 mm × 2 cm balloon (Bard, Tempe, Arizona). Post angioplasty angiography revealed a RPA stent fracture and an aorta to RPA fistula which was closed with a CP covered stent.

TABLE 1 Aorto-pulmonary fistulae

Patient	Diagnosis	Years from ASO	Prior interventions	Fistula	Stent fracture	Presentation	Management	Outcome
1	d-TGA	13	Prior RPA stent dilated 12 mm; LPA stent dilated 14 mm. Coronary artery stenting. (outside institution)	LPA	Yes	Congestive heart failure	CP 30 mm × 14 mm	AP fistula resolved
2	d-TGA/VSD	17	Prior RPA stenting with 16DS & PG 1910B. Prior LPA stenting with 16DS & PG2910B. RPA and LPA stents dilated to 16 mm	RPA	Yes	Angiography during procedure	CP 34 mm × 16 mm	AP fistula resolved
3	d-TGA/ASD	10	Prior RPA and LPA stenting with 16DS. Stents dilated to 14 mm	LPA	Yes	New continuous murmur	CP 35 mm × 14 mm	AP fistula resolved

Abbreviations: ASD, atrial septal defect; CP, P covered stent d-TGA, dextro-transposition of the great arteries; LPA, left pulmonary artery; PG, Palmaz Genesis, 16DS, DoubleStrut 16 mm stent; VSD, ventricular septal defect.

Patient 3: A 12-year-old underwent ASO in infancy. At 1 year, DoubleStrut 16 mm stents placed in the proximal branch pulmonary arteries. These stents were dilated to 10 mm at age three. They were redilated at age 10 to 14 mm. Two years later, she was noted to have a new continuous murmur with an AP fistula demonstrated by computerized tomography imaging. Catheterization confirmed the diagnosis and showed it was related to a fractured stent. The fistula was closed by implantation of a CP covered stent.

3.2 | Left main coronary artery compression

This patient previously had ASO followed 2 months later with revision of the LCA anastomosis and followed 7 years later by surgical left pulmonary artery patch-plasty. At age 11, the patient had stent implantation in the LPA. Two years later the stent was redilated causing compression of the LMCA and immediate bradycardia and left ventricular dysfunction. Urgent coronary stenting was performed using a 3.5 mm bare metal stent and function returned to normal (Table 2). Follow-up after 1-year showed normal LV function.

3.3 | High-risk situations (with intended pulmonary artery interventions)

We routinely perform coronary angiography and more recently 3D rotational aortography before interventions to delineate proximity of the coronary arteries and the bronchi to the targets of stenting. These evaluations have enabled recognition of high-risk situations, and have allowed us to modify intended procedures or to perform alternatives in order to prevent adverse events (Table 2).

Patient 1: A 14-year-old underwent ASO as a neonate. Twelve years later, the patient developed bilateral proximal pulmonary branch stenosis requiring intervention. Stenting of RPA was done successfully. Using 3D rotational angiography, we observed close proximity of the LMCA to the LPA stenosis and complete occlusion of the LMCA with a balloon testing in the LPA. We performed conservative serial angioplasty of the LPA stenosis instead of stenting. This was repeated 2 years later (Figure 3).

Patient 2: A 17-year-old underwent ASO and VSD closure as a neonate. Fourteen years later, the patient underwent cardiac catheterization for possible stenting of the supravalvar pulmonary stenosis, however, pulmonary angiography showed close proximity of LMCA to the narrowing. The patient was referred for surgical pulmonary valve replacement and pulmonary reconstruction.

Patient 3: A 13-year-old underwent ASO in the neonatal period. A LPA stent was implanted at age three. At age six, the patient had catheterization and LPA stent dilation was not performed because an aortogram suggested close proximity of the LMCA to the stent. Ten years later the patient had repeat catheterization because of the small LPA stent in addition to stenosis of the RPA branches. Stenting of upper and lower RPA branches was performed. However, 3D rotational angiography showed that the lower edge of LPA stent was in very close proximity to LMCA. Dilatation of the LPA stent was aborted (Figure 4).

TABLE 2 Coronary compression (CC) and high-risk situations with planned pulmonary artery interventions

Patient	Dx	Years from ASO	Coronary arrangement	Intended procedure	Actual procedure	Event	Imaging	Management
CC	d-TGA	13	Typical	LPA stenting	LPA stenting	LMCA compression, LV ischemia and dysfunction	Same day of procedure	Coronary stent
1(a)	d-TGA	12	Typical	RPA stenting; LPA stenting	RPA stenting, LPA balloon angioplasty	Occlusion of LMCA and aorta distortion during LPA balloon inflation	3DRA	LPA stent prohibited
1(b)	d-TGA	14	Typical	RPA stenting; LPA stenting	RPA stenting, LPA balloon angioplasty	Occlusion of LMCA and aorta distortion during LPA balloon inflation	Aortogram with simultaneous balloon inflation	Balloon dilations only
2	d-TGA VSD	17	Typical	MPA stenting	Diagnostic	Proximity of LMCA to RVOT	Coronary angiography during balloon sizing of RVOT	Referred for surgery
3(a)	d-TGA VSD	6	Typical	RPA balloon angioplasty; LPA stenting	RPA balloon angioplasty	Proximity of LMCA to LPA stent	Aortogram	LPA stent dilation aborted
3(b)	d-TGA VSD	13	Typical	LPA stenting	Diagnostic	Proximity of LMCA to LPA stent	3DRA	LPA stent dilation aborted. Future surgical management.
4	d-TGA VSD	12	Typical	Melody Valve	Melody Valve	Proximity of LMCA to RVOT	Coronary compression testing while balloon sizing of RVOT	Melody valve implanted at 18 mm instead of 20 mm

Stenting: New stent or dilation of prior stent.

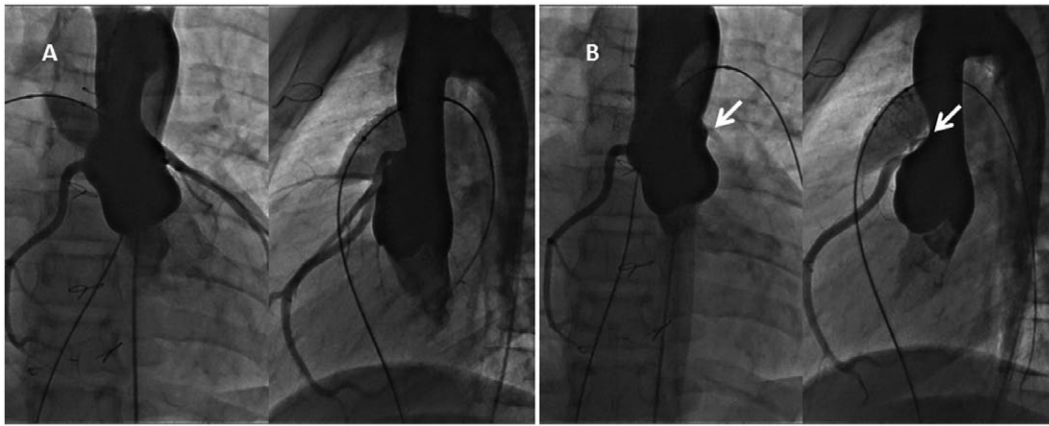


FIGURE 3 Fourteen-year-old male status post arterial switch operation as an infant and subsequent bilateral pulmonary artery branch stenosis and right pulmonary artery stenting. A, (PA and lateral) Aortogram with right pulmonary artery balloon inflation reveals patent coronary arteries. B, (PA and lateral) Aortogram with left pulmonary artery balloon inflation reveals occlusion of the left main coronary artery and aortic distortion (white arrow) prohibiting stent placement.

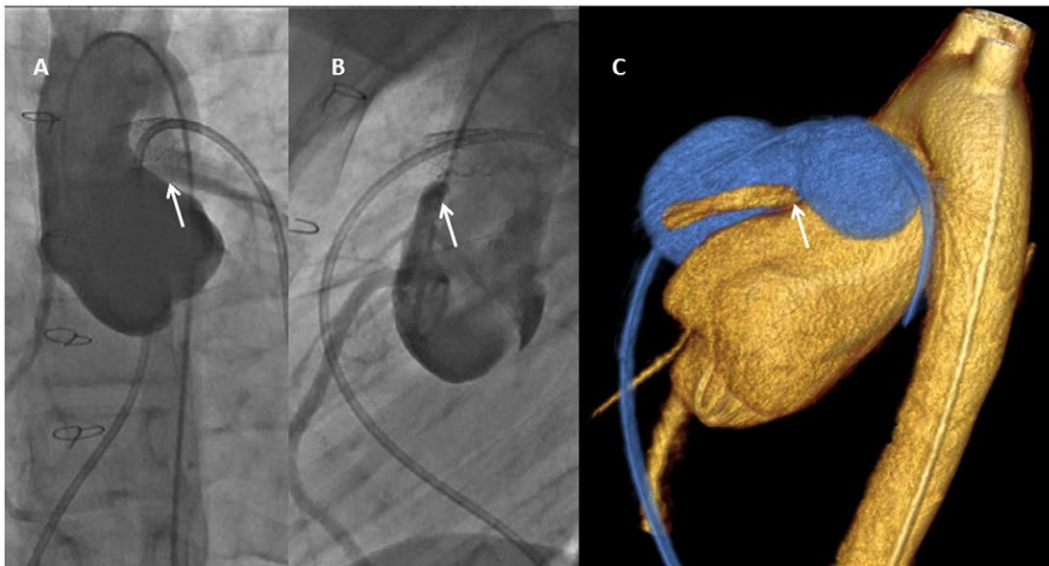


FIGURE 4 Thirteen-year-old male status post arterial switch operation as an infant and subsequent bilateral pulmonary artery branch stenosis and left pulmonary artery stenting. A, B, Aortogram in posterior-anterior and lateral views which show close proximity of the stent to the left main coronary artery (white arrow) which is at high-risk of compression if dilated. C, 3D rotational aortogram with simultaneous sizing balloon inflation in the left pulmonary artery allows evaluation from unconventional angles to confirm high-risk of left coronary (white arrow) compression.

Patient 4: A 22-year-old underwent an ASO with LeCompte maneuver and VSD closure as a neonate. This patient required transannular patch enlargement of the right ventricular outflow tract 4-years later. Several years later, right ventricular outflow tract stenosis recurred and the right ventricular outflow tract was revised and a mono-cusp valve was fashioned. Ten years later, the patient was brought to the catheterization laboratory for transcatheter pulmonary valve implantation because of recurrent stenosis and severe pulmonary regurgitation. Coronary compression testing was performed and showed proximity of the LCA to the right ventricular outflow tract requiring precise preimplantation stenting and

implantation of an 18 mm pulmonary valve instead of the intended 20 mm to avoid coronary compression.

4 | DISCUSSION

In this study, we reviewed catheterization procedures performed at our center in patients with d-TGA after ASO. We identified the types and frequency of interventions, complications, and high-risk situations, and we determined likely associated risk factors. In our series, 29 patients had 37 catheterization procedures,

which included angioplasty and/or stenting of pulmonary arteries.

There are few prior studies focused on catheter-based interventions in patients with d-TGA after the arterial switch operation and LeCompte maneuver. One recent report noted that 16 of 40 late interventional catheterization procedures were performed to address pulmonary artery stenosis in a series of 715 patients.⁴ In all cases balloon angioplasty was performed. In this study, no adverse events from angioplasty were reported, but procedural details were limited. Another report describes the requirement for intervention for supra-valvar pulmonary and left pulmonary artery stenosis in 29 of 103 patients.⁵ Balloon angioplasty was first line treatment in all patients, and stents were eventually placed in the pulmonary artery branches in 11 patients. No adverse events were reported. A third contemporary report describes 31 of 174 patients who developed pulmonary stenosis.⁶ Eleven patients underwent angioplasty, also apparently without notable adverse events. Finally, in an older report, authors describe angioplasty in 28 patients having 39 stenotic lesions.⁷ Adverse events included one ruptured vessel and three pseudo-aneurysms.

Studies on angioplasty and stenting of pulmonary arteries suggest that in general morbidity and mortality are relatively low. Data from the Congenital Cardiac Catheterization Project on Outcomes Registry describing both pulmonary artery angioplasty and stenting procedures reported a major adverse event or death rate of 2.9% (38/1315 patients).⁸ Data from the National Cardiovascular Data Registry's IMPACT Registry reported a major adverse event and death rate in proximal pulmonary branch stenosis stenting of 2.0% (5/249 patients) including one patient who had unplanned surgery after catheterization and two patient deaths during the episode of care related to the stent procedure.⁹ In a later report from IMPACT, the authors calculated a major adverse event or death rate of 9% in 1183 patients who underwent pulmonary artery stenting. In this report, it was observed that major adverse events or death were most associated with specific patient characteristics (less than 4 kg, single ventricle, emergency stenting procedure).¹⁰ This study could not have identified the LeCompte maneuver as a risk factor, because it is not a data element in the study's analysis.

Our review suggests that interventions on pulmonary branch stenosis in patients who had ASO and the LeCompte maneuver were undertaken for indications typical of most patients with postoperative pulmonary artery stenosis. In our study, none of the patients had risk characteristics identified in the second IMPACT study cited earlier. Nevertheless, if we consider only the catheterization procedures involving angioplasty and/or stenting of pulmonary arteries, the incidence of major complications or death (using definitions similar to those in the registry reports) was 14% (5/37 procedures) in our series. If we consider only stent procedures, the rate of major complications was 23% (5/21 procedures). High-risk situations were identified in an additional 16% (6/37) and planned procedures were aborted. It is notable, however, that adverse events were seen in patients who had previous uneventful interventions on the pulmonary branches. The rates cited above were observed in patients who were

many years post ASO, and simply were rates experienced over a 10-year period among patients referred for intervention. Nevertheless, our series suggests that intervention in this patient population is high-risk. Clearly, an appropriate risk/benefit calculation should be performed prior to performing interventions on pulmonary arteries in this patient population.

Among the major adverse events we encountered were three cases with AP fistulae following pulmonary arteries branch stenting or stent dilation. Stent fractures were detected in all cases and restenting had been performed in one without noting a fistula.

In patients after ASO with LeCompte maneuver, there are only a few case reports of AP fistula following pulmonary angioplasty or angioplasty and stenting. The earliest report was in 1994 by Preminger and Lock after LPA angioplasty.¹¹ Eleven additional cases have been reported.¹²⁻²⁰ Five were after one or more angioplasties, and six cases occurred after angioplasty and stent implantation. Of the stented cases, none were noted to have stent disruptions or fractures. Two of the 12 fistulae were very small and were not treated. Ten were closed: four with covered stents,^{11,14,17,20} three with surgery,^{12,13,19} two with aortic endografts^{16,18} and one with a device.¹⁵

In context of these few reports, we find our experience illuminating. We believe that factors in our cases leading eventually to fistulae were the LeCompte arrangement of the stenotic pulmonary branches, the use of bare metal stents at target sites along the straddling arteries, redilation of the stents to larger than recommended diameters, and finally stent fractures. It is also notable that the fistula patients all had stent implantation in both pulmonary artery branches. Perhaps this further limited the compliance of the vessels to accommodate the aorta. Consistent with highly associated risk factors in a previous report,²¹ we believe that forces on the stents from the adjacent pulsatile high-pressure aorta, and larger than recommended stent diameters were factors leading to fractures and sharp broken struts. Fistulae occurred near the sites of the stent breaks presumably from the pressure and motion of the aorta and other cardiac structures. While we think that specific stent type or bilateral stenting may be specific risk factors, we realize our data is limited and anecdotal. We were able to close the fistulae using covered stents, however we await longer-term follow up to determine if the process outlined above may repeat itself.

Our review also identified one patient who had LMCA compression caused by stent implantation in the LPA. The patient presented in heart failure secondary to severe LV dysfunction, which necessitated urgent coronary artery stenting. As in reports by Hamzeh and Van Gameren,^{22,23} LMCA compression may occur after pulmonary artery stenting from the mass effect of the stents if the coronary artery origin or course is close to the target of stenting. Given the necessity to translocate the coronary arteries and the LeCompte, coronary arteries may be translocated to sites in the neo-aorta adjacent to sites in the pulmonary artery or pulmonary branches, which become stenotic. This sets up potential high-risk situations if operators do not fully appreciate the proximity.

We also encountered six significant high-risk situations (16%) among additional patients having planned pulmonary artery

interventions. In all cases there was threatened LMCA compression due to the close proximity to the pulmonary arteries during planned stenting procedures. These cases occurred later in our experience and benefited from our contemporary approach for evaluation of the coronary arteries prior to percutaneous pulmonary valve implantation. We used compression testing to demonstrate the effects of a stent implantation or 3D rotational angiography to identify close proximity of the coronary arteries to sites of pulmonary stenosis. Taking a conservative approach to avoid major complications, we abandoned or modified our planned interventions in patients with either positive compression tests or with close proximity of stenosis to the targets of stent implants. Notably, our observations are confirmed by a recent report using similar methods in a sub-group of patients with TGA after ASO undergoing evaluation for transcatheter pulmonary valve implantation. In this study, 19% had risk of coronary artery compression and transcatheter valve implantation was abandoned.²⁴

Given our experience, we suggest that if pulmonary artery interventions are being considered in this population as the principle indication for catheterization, a preprocedural advanced imaging study to evaluate the proximity of the coronary arteries is helpful. In addition, during the procedure, rotational angiography with balloon sizing of the stenosis with coronary compression testing should be performed if there are concerns.

5 | CONCLUSION

This review suggests that percutaneous intervention on pulmonary artery stenosis after ASO has high-risk and should be undertaken advisedly. Prior thorough evaluation of coronary arteries is mandatory as coronary reimplantation sites may be adjacent to sites of pulmonary artery stenosis. Furthermore, if pulmonary artery stent implantation or stent redilation is contemplated, the risk of stent fracture and possible AP fistula should be recognized. Primary use of reinforced covered stents should be considered.

AUTHOR CONTRIBUTIONS

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