Feasibility of combining additional interventions to facilitate hemodynamics during Transcatheter Fontan procedure without prior surgical preparation

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ABSTRACT

Transcatheter Fontan is an alternative palliation in severely hypoxic univentricular hearts at high-risk for Fontan surgery. While sternotomy, prolonged intubation, and positive-pressure ventilation impede passive systemic venous return, transcatheter Fontan allows early extubation and mobilization to promote inspiratory venous return, additionally aided by the skeletal muscle pump. Following puncture of the pulmonary artery (PA) and atrium through the transverse pericardial sinus, a long-covered stent bridges the inferior vena cava to the PA. Concurrent interventions create wide inter-atrial communication, interrupt competitive aortopulmonary or antegrade flows, and relieve PA stenosis. This case series details such interventions to facilitate optimal hemodynamics after establishing neo-Fontan circulation.

Keywords: Antegrade pulmonary blood flow, atrial septostomy, bidirectional Glenn surgery, Blalock–Taussig shunt, covered stent, univentricular heart

INTRODUCTION

Univentricular heart diseases often have poor survival, unless surgically intervened.[1] The initial surgical plan typically includes a modified Blalock–Taussig shunt (mBTS) or ductal stent to improve pulmonary blood flow or pulmonary artery (PA) banding to restrict it. After a second stage bidirectional Glenn (BDG) shunt, the final palliation is Fontan completion with facilitatory steps including (i) closure of antegrade pulmonary blood flow, patent ductus arteriosus (PDA), mBTS or aortopulmonary collaterals, (ii) repair of PA stenosis and regurgitant atrioventricular valves, (iii) widen atrial communications and bulboventricular foramen and (iv) closure of veno-venous collaterals directly draining into the coronary sinus or pulmonary veins. When patient age, PA pressure, and size, pulmonary vascular resistance, and ventricular function are not ideal, surgery carries a high risk.[2] Transcatheter Fontan completion is an evolving alternative in such patients.[3,4] This case series illustrates the feasible concurrent procedures during a transcatheter Fontan completion to facilitate the hemodynamics.

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CASE REPORTS

Case 1

A 15-year-old male with Duchenne muscular dystrophy underwent left thoracotomy mBTS at 6 months for

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double outlet right ventricle, nonroutable ventricular septal defect (VSD), severe pulmonary stenosis (PS), and d-malposed aorta. During an off-pump BDG at 1-year of age, dense mediastinal adhesions around the mBTS posed surgical challenges for dissection around the shunt and hence was left behind. Atrial septectomy was not performed as the echocardiogram showed a small but adequate interatrial communication.. The distal PA was banded. Oxygen saturation were reasonably maintained by this "super Glenn" circulation combining mBTS and BDG, and parents did not seek medical attention.[5] Diagnosis of myopathy due to Duchenne muscular dystrophy also deterred the family from seeking further medical attention towards completion of Fontan surgery. When saturations dropped 14 years later to 79%, PA pressure on catheterization was 18 mmHg, transpulmonary gradient was 6 mmHg, and ventricular end-diastolic pressure was 15 mmHg. Nakata index of 360 mm^2/m^2 and PA arborization were adequate. Right atrial angiogram showed an intact atrial septum [Figure 1]. Echocardiogram confirmed an intact atrial septum, normal ventricular systolic function, and absence of atrio-ventricular valve regurgitation. As advanced age, myopathy, high PA pressures, and filling pressures posed high risk, we preferred the transcatheter Fontan to allow early extubation and mobilization.

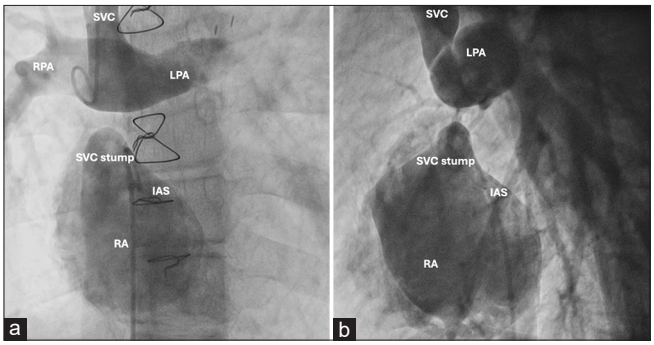


Figure 1: Simultaneous superior vena caval and right atrial angiogram in anteroposterior (a) and lateral view (b) demonstrates a good-sized right pulmonary artery and left pulmonary artery (LPA) and intact atrial septum. Posthilar LPA flows are reduced due to competitive Blalock–Taussig shunt flows. RA: Right atrial, RPA: Right pulmonary artery, LPA: Left pulmonary artery, IAS: Intact atrial septum, SVC: Superior vena cava

After right jugular and bilateral femoral venous access, Brockenbrough needle puncture of the atrial septum, and serial balloon dilations culminating in a 30 mm × 4 cm Z-med balloon (Numed Inc, Hopkinton, NY) created an adequate septostomy [Figure 2]. The PA band was closed from the right jugular vein with a 6-4 Amplatzer duct occluder II (Abbott Medical, Plymouth, MN) to stop the antegrade flow [Figure 3].

A transjugular Brockenbrough needle advanced through an 8F Mullins sheath (Cook Medical, Bloomington, IN) punctured the floor of the right PA and roof of left atrium through the transverse pericardial sinus using monopolar diathermy in cutting mode at 50W delivered for 2-3 seconds [Figure 4]. A guidewire through this Mullins sheath was snared from the femoral venous sheath to form a venovenous circuit. As a single 12 cm long covered stent to bridge the distance between the hepatic veins to the right PA was unavailable, it was decided to combine a covered stent at the cranial PA end and a hybrid stent with a 16 mm uncovered part for the caudal inferior vena cava (IVC) end.[5]

A long 20F braided straight Performer sheath (Cook Medical, Bloomington, IN) was advanced from the right femoral vein over this guidewire circuit into the right superior vena cava (SVC) through the punctured zone. The cranial end of a 79 mm long covered Zephyr XL (Sahajanand Laser Technologies, Gujarat, India) mounted on a 22 mm × 8 cm Z-med balloon (Numed Corporation, Hopkinton, NY) was exposed beyond the tip of the sheath in SVC and expanded near the floor of the right PA. A venous collateral from the SVC to the right upper pulmonary vein identified on right jugular venogram was left to act as a Fontan fenestration. After unsheathing and expanding the remaining stent, a second 80 mm long hybrid covered Zephyr stent was caudally overlapped using the same balloon [Figure 5]. The caudal uncovered part astride the hepatic veins in IVC was postdilated with a 28 mm × 4 cm Z-med balloon.

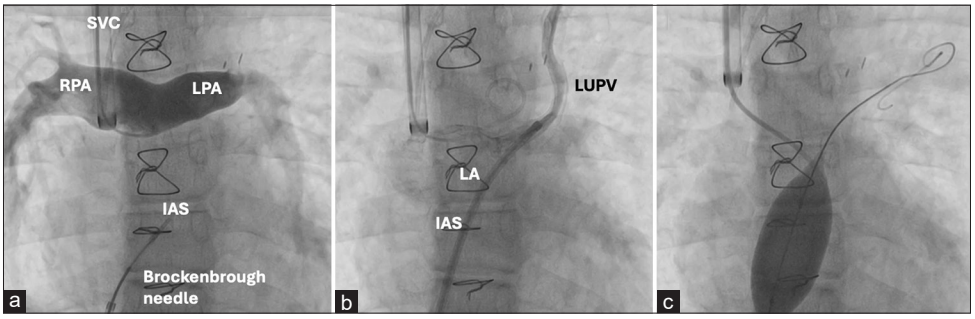


Figure 2: Angiogram (a) from a large jugular sheath in right superior vena cava delineates right pulmonary artery and left pulmonary artery, while a transseptal Brockenbrough needle punctures the interatrial septum to get a long sheath (b) into the left upper pulmonary vein through the left atrium. Balloon septostomy with a large balloon provides an atrial communication (c). RPA: Right pulmonary artery, LPA: Left pulmonary artery, IAS: Intact atrial septum, SVC: Superior vena cava, LA: Left atrium, LUPV: Left upper pulmonary vein

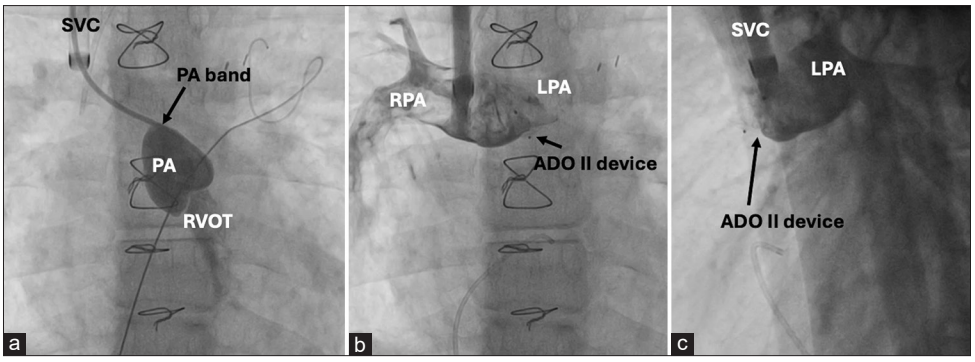


Figure 3: The pulmonary artery band crossed by a catheter (a) from superior vena cava demonstrates pulmonary valve and right ventricular outflow tract. Closure of the banded site with an occluder device is shown in anteroposterior (b) and lateral views (c). PA: Pulmonary artery, RPA: Right pulmonary artery, LPA: Left pulmonary artery, SVC: Superior vena cava, RVOT: Right ventricular outflow tract, ADO: Amplatzer duct occluder II device (Abbott Medical, Plymouth, MN)

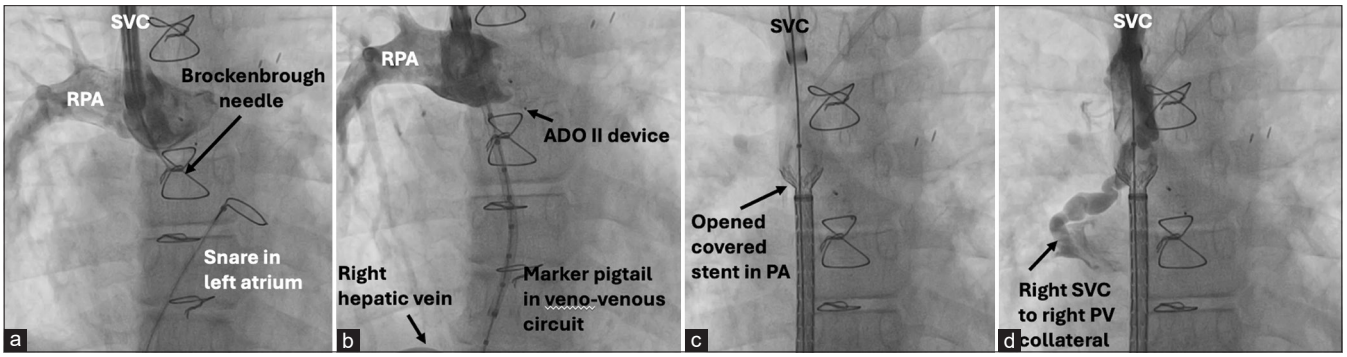


Figure 4: A transjugular Brockenbrough needle (a) from superior vena cava (SVC) punctures the floor of the right pulmonary artery (RPA) towards a snare loop in the left atrium to create a femoro-jugular guidewire circuit. Marker pigtail measures distance (b) between right hepatic vein and RPA. The cranial struts of a long-covered stent are expanded in the SVC and pulled towards the floor of the RPA (c). A venovenous collateral from SVC to right upper pulmonary vein on jugular sheath angiogram (d) will act as a Fontan fenestration. SVC: Superior vena cava, RPA: Right pulmonary artery, ADO: Amplatzer duct occluder II device (Abbott Medical, Plymouth, MN)

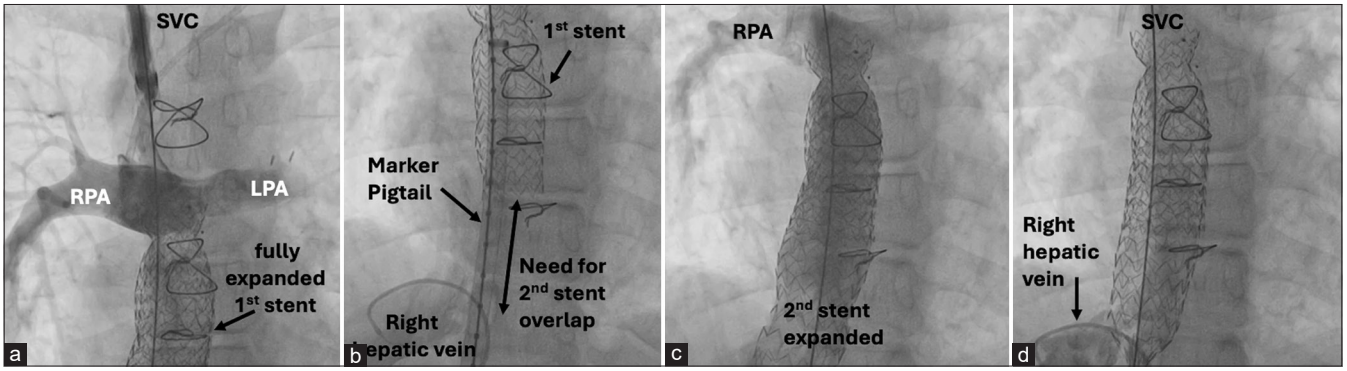


Figure 5: After fully expanding the first 79 mm covered stent (a), marker pigtail remeasures the distance from lower end of stent to hepatic vein (b). After expanding a second hybrid stent caudally (c), right hepatic vein angiogram (d) shows filling of the right pulmonary artery and left pulmonary artery. superior vena cava. RPA: Right pulmonary artery, LPA: Left pulmonary artery, SVC: Superior vena cava

The PA end of the left mBTS was closed through jugular access by a 6 mm Amplatzer vascular plug II (Abbott Medical, Plymouth, MN) to arrest competitive flows [Figure 6]. Postprocedural Fontan pressure was 16 mmHg. The patient was extubated within 2 h and ambulated within 8 h. At a 6-month follow-up on coumadin, frusemide, deflazocort, and sildenafil, oxygen saturations were 94% due to the venous collateral from the SVC to the right upper pulmonary vein [Figure 4].

Case 2

A 10-year-old male with pulmonary atresia, intact ventricular septum underwent neonatal valve perforation and balloon dilation. As the tricuspid valve Z-score was < -2, right BDG was added at one year, along with a PA

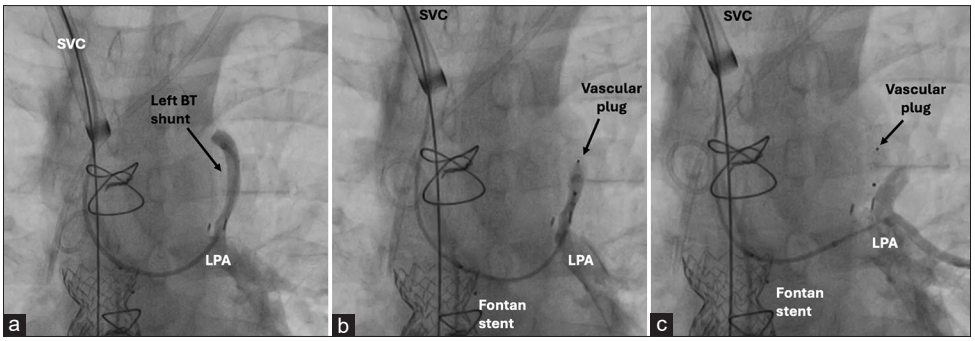


Figure 6: Catheter advanced from superior vena cava into the left pulmonary artery (LPA), cannulates the left Blalock–Taussig shunt (a) and aids its closure by a vascular plug (b). Subsequent LPA angiogram (c) shows adequate hilar arborization. LPA: Left pulmonary artery, BT: Blalock–Taussig, SVC: Superior vena cava

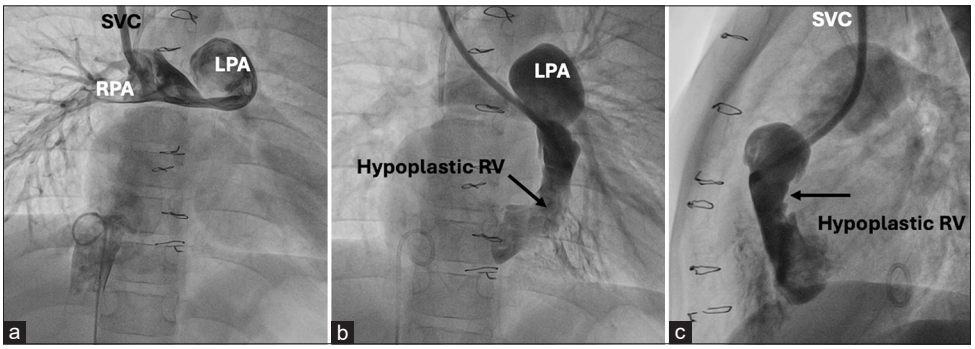


Figure 7: Superior vena caval angiogram (a) shows Glenn shunt with mild ostial narrowing of right pulmonary artery and large left pulmonary artery. A sheath angiogram through the pulmonary artery shows antegrade flows from a hypoplastic right ventricle on anteroposterior (b) and lateral (c) views. RPA: Right pulmonary artery, LPA: Left pulmonary artery, SVC: Superior vena cava, RV: Right ventricle

band to restrict antegrade flow. When Glenn pressure was 16 mmHg and left ventricular end-diastolic pressure was 14 mmHg at 10 years of age, the heart team opted for transcatheter Fontan completion. Ostia of the right PA, though narrowed, did not show any gradient [Figure 7].

After jugular and femoral vein access, the PA band was closed using a 14–12 Konar multifunction occluder (Lifetech Scientific Co, Shenzhen, PRC) through a 7F sheath [Figure 8]. After an electrosurgical puncture of the floor of the right PA and roof of the left atrium, a venovenous circuit was established between the right jugular vein and right femoral vein to advance a 20F long Performer sheath through the puncture zone from the right groin. Cranial end of a 79 mm Zephyr covered stent mounted on a 22 mm × 8 cm Zmed balloon was expanded within the right PA. After unsheathing and full expansion of the entire stent, the IVC end was postdilated with a large 28 mm balloon [Figure 9]. The cranial right PA end of the stent was flared using two 12 mm × 4 cm Mustang balloons (Boston Scientific, Marlborough, MA) after advancing two stiff guidewires from the right groin sheath into both pulmonary arteries [Figure 10]. The final angiogram showed a good seal of the lower end of the stent without any residual flows. The saturations improved to 98%. Even though the PA pressure increased from 16 mmHg to 18 mmHg, the heart team planned to manage the patient with diuretic infusions.

Over the next few days, high Fontan pressures led to liver dysfunction and reduced urine output. On a re-catheterization 2 weeks after the transcatheter Fontan procedure, the fabric of the covered stent was fenestrated using a Brockenbrough needle, resulting in a drop of oxygen saturation to 70%. When renal and liver functions recovered over the next 1 month, he underwent a stent overlap for the lower end using an 8 cm hybrid stent, that was postdilated to 34 mm using a sizing balloon. The oxygen saturation improved to 88% at 1-month follow-up, while he was still receiving diuretics, coumadin, and sildenafil.

Case 3

A 10-year-old girl with an unbalanced atrioventricular canal defect (u-AVCD), PS underwent BDG in infancy. Sildenafil and frusemide were started at 10 years when PA pressure was 18 mmHg and transpulmonary gradient was 7 mmHg. With no hemodynamic improvement after 2 years, the heart team opted for transcatheter

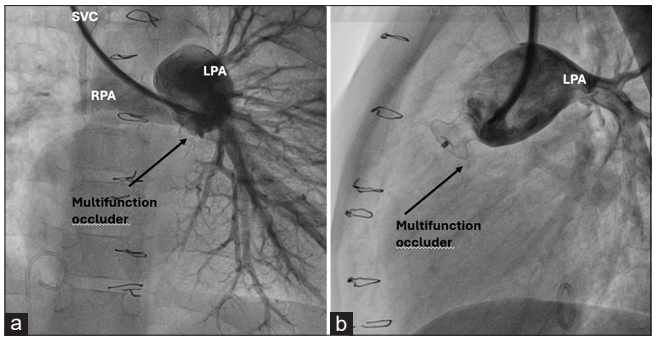


Figure 8: Antegrade pulmonary blood flow is closed by the multifunction occluder device, while a superior vena caval angiogram in anteroposterior (a) and lateral (b) views shows right and left pulmonary arteries. RPA: Right pulmonary artery, LPA: Left pulmonary artery, SVC: Superior vena cava

Fontan completion as saturation was 78%. After jugular and femoral vein access, the floor of the right PA was punctured using electrosurgery, a venous circuit was made, and two overlapping 79 mm Zephyr covered stents were deployed to bridge the IVC towards the PA. The contrast stasis within the conduit and increase of Fontan pressures to 20 mmHg indicated the inability of the pulmonary vascular bed to tolerate the entire caval return and warranted an immediate, reliable fenestration. Transjugular Brockenbrough needle punctured the stent fabric, to advance a 7F sheath and deploy an 8 mm × 2 cm Formula 535 stent (Cook Medical, Hopkinton, NY) to act as a fenestration [Figure 11]. At 18-month follow-up, fenestration was patent to maintain saturations at 93% while receiving frusemide and coumadin.

Case 4

A 14-year-old male underwent BDG at 1 year for Tricuspid atresia, restrictive VSD, PS, and restrictive PDA. PA pressure on preFontan catheterization was 17 mmHg, and transpulmonary gradient was 7 mmHg. The heart team preferred transcatheter Fontan completion. An arterial catheter was advanced through the PDA and narrowed pulmonary valve into the hypoplastic right ventricle, closing the antegrade flow with a 6-6 Amplatzer duct occluder II and PDA with a 5-4 Piccolo device (Abbott Medical, Plymouth, MN). Brockenbrough needle through jugular access punctured the right PA to achieve a venovenous circuit [Figure 12]. A single 10 cm covered Zephyr stent was deployed from the right PA to the IVC using a 22 mm × 10 cm balloon, and the lower end was expanded with a 30 mm × 4 cm balloon. As the initial PA pressure was 17 mmHg, a longitudinal 5 mm slit was made with a surgical blade on the stent fabric before crimping to the balloon to expand and act as a fenestration. The final Fontan pressure was 17 mmHg. On a 1-month follow-up, the oxygen saturation was 88% while receiving frusemide, sildenafil, and coumadin.

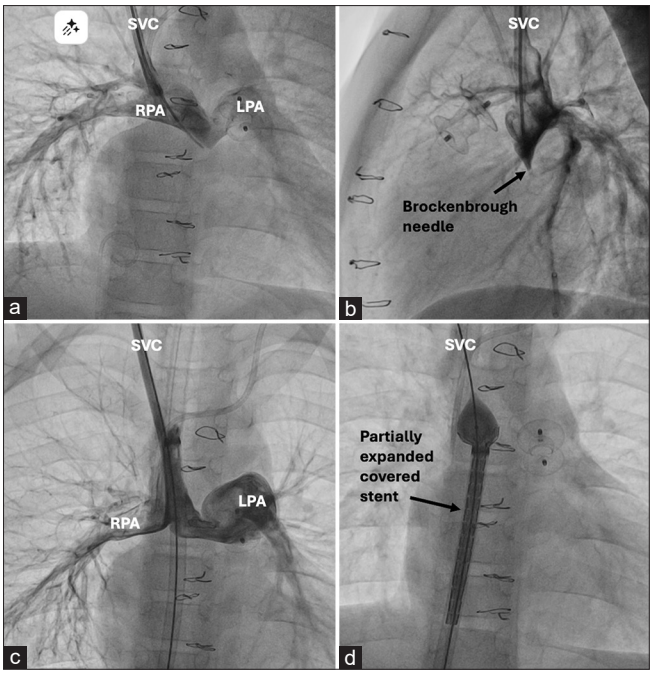


Figure 9: Transjugular Brockenbrough needle from superior vena cava (SVC) punctures the floor of right pulmonary artery (RPA) on anteroposterior (a) and lateral (b) views to achieve a venovenous guidewire circuit (c). The cranial end of the covered stent is expanded in the SVC and pulled up to the floor of the RPA (d). LPA: Left pulmonary artery

Case 5

A 30-year-old male with tricuspid atresia and restrictive VSD underwent BDG at 4 years of age, along with pericardial patch enlargement of the left PA ductal site stenosis. Lost to follow-up for the next 16 years, pre-Fontan catheterization showed complete occlusion of the left PA that was recanalized by stent angioplasty. Even though a re-catheterization after 1 year showed acceptable PA pressures of 14 mmHg, advanced age and focal segmental glomerulosclerosis-related azotemia did not suit Fontan surgery. He presented with oxygen saturations of 78% hematocrit levels of 78% at 30 years of age. Following electrosurgical puncture of the floor of the right PA and the roof of the atrium to establish a venovenous circuit, two long covered stents completed transcatheter Fontan completion. The left PA stent was redilated with a 14 mm Atlas gold balloon (Bard Vascular, Tempe, AZ) to optimize Fontan flows to both lungs [Figure 13]. At 1-year follow-up on coumadin and losartan for proteinuria, his symptoms improved considerably.

Case 6

A 22-year-old male with right isomerism, dextrocardia, u-AVCD, dominant right ventricle with previous left BDG at 2 years, presented after 20 years with saturations of 70%, hematocrit of 71%, and uric acid of 10.5 mg/dl.

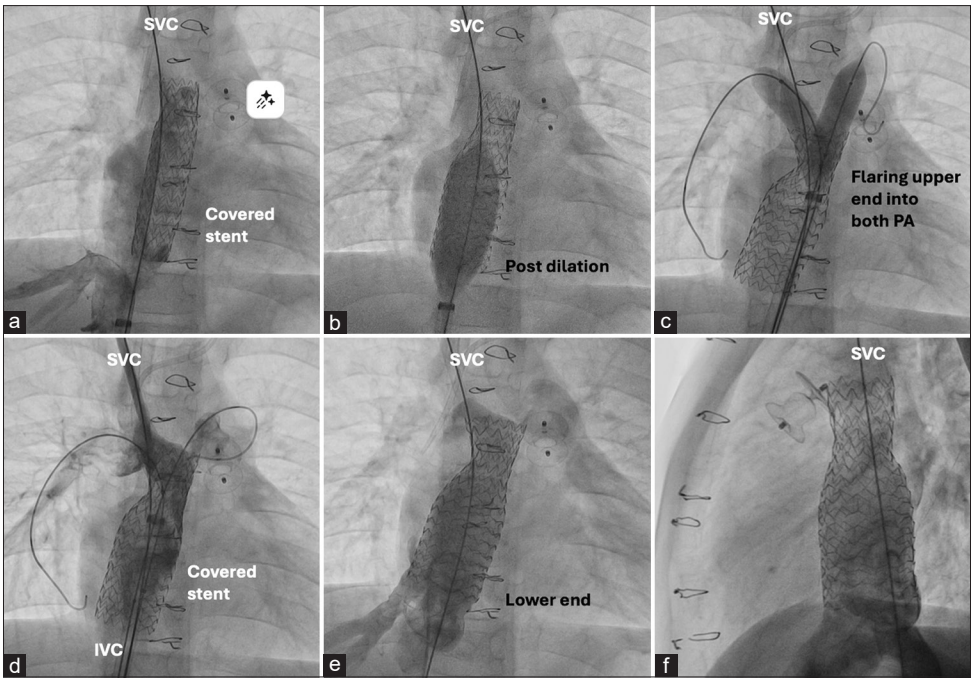


Figure 10: The caudal end of the fully expanded covered stent is not fully opposed to the inferior vena cava (IVC) on hepatic vein angiogram (a) before postdilation of the lower end with a large balloon (b). The upper end of the covered stent is flared by two balloons advanced into the branch pulmonary artery to provide a tulip configuration (c). Final superior vena caval angiogram (d) and IVC angiogram in anteroposterior (e) and lateral (f) views show unobstructed Fontan flows. SVC: Superior vena cava, IVC: Inferior vena cava

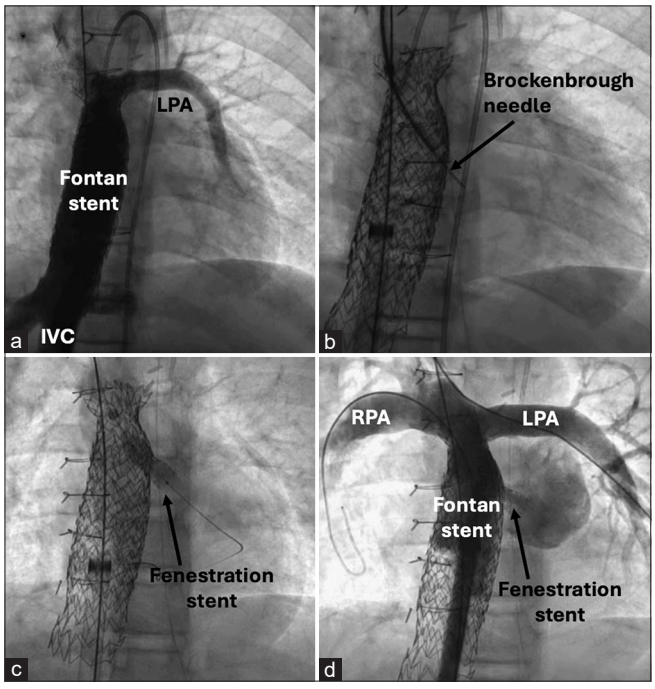


Figure 11: High pulmonary artery pressures make stagnation of contrast during inferior vena caval angiogram after transcatheter Fontan completion (a). Brockenbrough needle from jugular vein punctures the stent fabric (b) to allow a small 8mm stent (c) that fenestrates the Fontan circuit (d). RPA: Right pulmonary artery, LPA: Left pulmonary artery, IVC: Inferior vena cava

With PA pressures of 13 mmHg and a transpulmonary gradient of 8 mmHg, indexed pulmonary vascular resistance was 4.2 wood units. Multiple aortopulmonary collaterals from aortic arch branches were occluded with coils, followed by the establishment of a covered stent connection from IVC to left PA [Figure 14]. Even though the PA pressures increased only to 16 mmHg, the heart team planned a prophylactic fenestration. The stent fabric was punctured with a Brockenbrough needle and dilated with a 6 mm balloon, resulting in exit saturations of 90% [Figure 15]. During a 15-month follow-up, hematocrit improved to 55%, and symptoms improved considerably.

DISCUSSION

Transcatheter Fontan completion involves four crucial procedural steps.[6] First step punctures the floor of the PA and roof of the atrium through the transverse sinus, assisted by electrosurgery. Surgical adhesions around prior BDG offer safety from a mediastinal bleed. The distance between PA and hepatic veins on the guidewire circuit formed during the second step guides the selection of the stent length. A single long covered stent or two overlapping stents bridge the IVC to PA in the third step. Uncovered end of a hybrid stent for the caudal hepatic venous segment of IVC allows drainage of hepatic veins through the open side-struts. The utility of this hybrid covered stent is previously reported in different variations of sinus venosus defects.[5] To secure the superior PA end of the covered stent, the

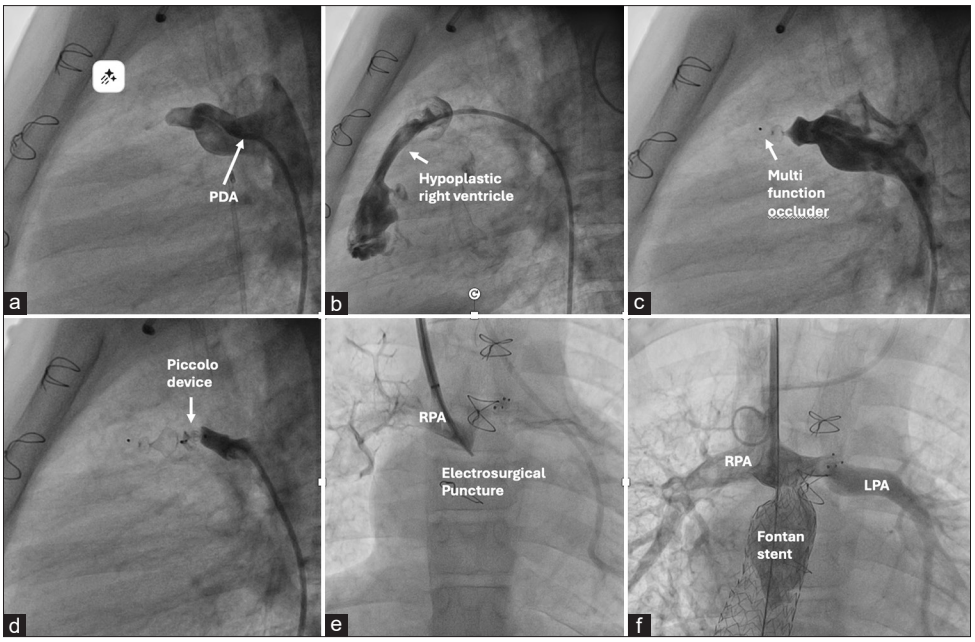


Figure 12: Lateral-view aortogram (a) shows patent ductus arteriosus (PDA). Advancing catheter into the hypoplastic right ventricle (b) allowed closure with a multi-function occluder (c) and subsequent PDA closure (d) with Piccolo device. Following electrosurgical puncture (e) of floor of right pulmonary artery, transcatheter Fontan circuit (f) is established. PDA: Patent ductus arteriosus, RPA: Right pulmonary artery, LPA: Left pulmonary artery

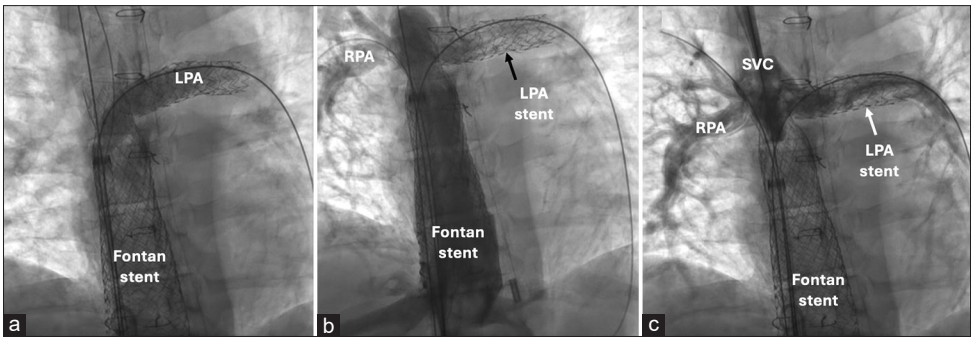


Figure 13: Left pulmonary artery (LPA) stent is postdilated following transcatheter Fontan (a). Angiogram from Fontan stent (b) and superior venacava show contrast flows (c) into both right pulmonary artery and LPA. RPA: Right pulmonary artery, LPA: Left pulmonary artery, SVC: Superior vena cava

first 1 cm is often expanded within the SVC to pull it against the floor of the PA. This step ensures stent stability. The last step flares the upper end using two balloons advanced into the right and left PA, creating a tulip-like configuration at the upper end to optimize the best flows.[6]

Although a well-performed surgical Fontan provides the best flow dynamics, surgery carries a distinct risk in advanced age, ventricular dysfunction, high PA pressures, and transpulmonary gradients. In such hypoxic patients, transcatheter Fontan offers a less invasive opportunity to improve oxygenation. A nonsurgical procedure without surgical chest incisions allows early postprocedural extubation. Early spontaneous breathing improves the inspiratory pull of venacaval blood to the chest. As there are no chest drains, patients can be mobilized out of bed within a day. Skeletal leg muscles pump the venous blood towards the lungs during early ambulation after a Transcatheter Fontan procedure.

The interventions described above needed multiple additional steps, such as (i) atrial septostomy to equalize interatrial pressures, (ii) antegrade flow closure using devices across the PA band, (iii) mBTS closure, (iv) PDA closure, (v) aortopulmonary collateral closure, (vi) PA stent enlargement, and (vii) fenestration creation by puncturing the stent fabric.[7,8] Even though these concurrent procedures are more easily achieved during surgical Fontan completion, the additional interventional steps need proper procedural planning.

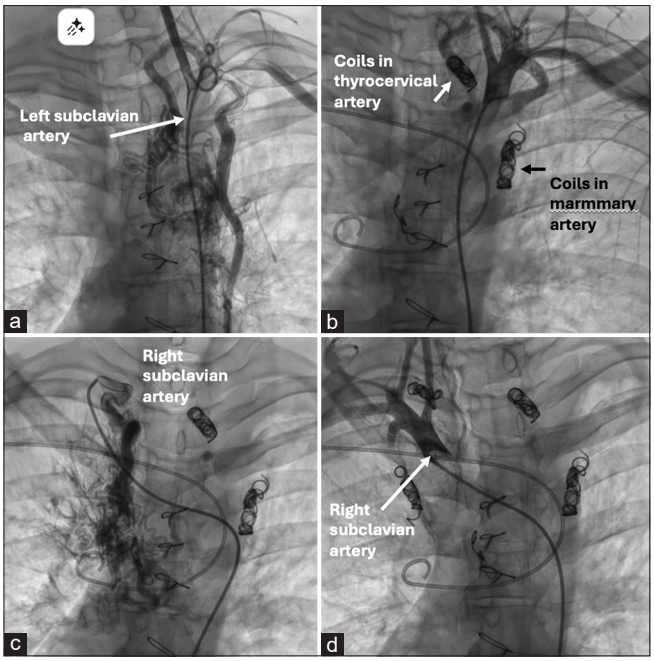


Figure 14: Angiogram in left subclavian artery shows collaterals (a) closed with coils (b). Collaterals from right subclavian artery (c) is also closed with coils (d)

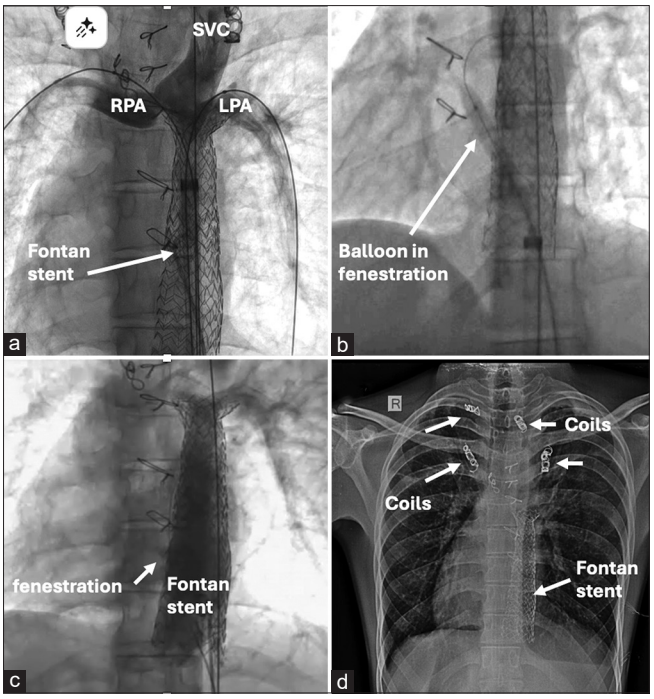


Figure 15: Superior vena caval (SVC) angiogram after transcatheter Fontan completion (a) shows right (RPA) and left (LPA) pulmonary arteries. Puncture of the stent fabric (b) creates a fenestration seen on angiogram (c) Xray chest after 15 months (d) shows the coils and Fontan stent

Other concurrent procedures that may be needed in some of the transcatheter Fontan procedures include *de novo* PA stenting and closure of very large venous collaterals, decompressing the SVC into the pulmonary veins or coronary sinus.[9-11] Optimization of Fontan hemodynamics is vital to prevent competitive PA flows and maintain low PA pressures.

A fenestration offers a safety net after any Fontan procedure, but the downside includes postprocedural hypoxia, paradoxical embolism, and later persisting polycythemia. We chose different modes of fenestrations in these patients. A small slit incision made in the covered stent before crimping to the balloon can be adopted when fenestration is considered mandatory. In other patients, when poststent angiogram shows contrast stasis or high Fontan pressures, the fabric can be easily fenestrated with a Brockenbrough needle and dilated with a small balloon with or without a stent.

In an ideal patient who satisfies all criteria for a successful Fontan candidacy, an elegantly placed smooth polytetrafluoroethylene surgical conduit with an offset from the Glenn flows provides the best computational flow dynamics for the passive venous flows with minimal energy loss. However, in extremely hypoxic patients who fail these criteria, surgical palliations such as the addition of a super Glenn or creation of a cubital arteriovenous fistula are equally morbid and very rarely performed. In such unfortunate patients, transcatheter Fontan is a safer alternative to the above-mentioned palliations. Unlike surgical conduits that are uniform in diameter, transcatheter Fontan stents are often narrower at the pulmonary end than the caudal end. Long follow-up should focus on the impact of this varying diameter on the passive venous flows. Given the metallic artifacts associated with these stents, magnetic resonance imaging is unlikely to offer any meaningful information on the flows and energy loss.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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