Case Report

Acute Thrombosis During Ductus Arteriosus Stenting Successfully Treated with Balloon Angioplasty and In Situ Alteplase Infusion: Case Report

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Introduction

Tricuspid atresia with pulmonary atresia is a rare congenital heart defect, representing about 1% to 3% of all congenital heart diseases.[1] Tricuspid atresia is subdivided into type I (normal position of the great arteries) and type II (transposition of the great arteries), with type 1A characterized by the presence of associated pulmonary atresia or stenosis.[2] Pulmonary blood flow in these patients is dependent on the patency of the ductus arteriosus, making its maintenance essential in the neonatal period.

The treatment of tricuspid atresia follows a staged surgical approach, aiming for a Fontan circulation as the final goal. In the neonatal period, the focus is to ensure adequate pulmonary blood flow through hemodynamic or surgical interventions, including systemic-pulmonary shunts or ductal interventions.[3]

This case report describes a newborn with type 1A tricuspid atresia who underwent a hemodynamic intervention with stent implantation in the ductus arteriosus, discussing technical aspects of the procedure, complications, and clinical evolution.

Case report

A term male newborn weighing 2,920 g was transferred at 6 days of life to a cardiac intensive care unit. The patient was hemodynamically stable, with oxygen saturation of 85% in room air, and was receiving a continuous intravenous infusion of prostaglandin at 0.01 mcg/kg/min. Transthoracic echocardiography demonstrated tricuspid valve atresia, pulmonary valve atresia, a hypoplastic right ventricle, the aorta arising from the left ventricle, and a tortuous ductus arteriosus (type 1A tricuspid atresia) (Figure 1). Chest computed tomography angiography showed a tortuous ductus arteriosus and stenosis at the origin of the left pulmonary artery (Figure 2). The patient also had a multicystic dysplastic left kidney. After multidisciplinary discussion, ductal stenting was chosen as a palliative procedure in the neonatal period.

Keywords

Congenital Heart Defects; Tricuspid Atresia; Ductus Arteriosus; Percutaneous Coronary Intervention; Thrombosis

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Interventional procedure

Due to the angle between the aortic end of the ductus arteriosus and the aortic arch, the right carotid artery was chosen as the access route. To improve catheter and guidewire manipulation and ergonomics, the patient was positioned in reverse orientation on the hemodynamic table (Figure 3).

The right carotid artery was punctured under ultrasound guidance, and a 5F slender transradial introducer was placed. Intravenous heparin (100 IU/kg) and prophylactic cefazolin (50 mg/kg) were administered. Left heart catheterization and cineangiography confirmed the tortuous ductal anatomy and the stenosis at the origin of the left pulmonary artery (proximal to the ductal insertion). A 0.014″ Balance Heavy Weight (BHW) guidewire was placed in the left pulmonary artery with microcatheter support. A Mini Trek 2 × 8 mm balloon catheter was positioned in the ductal trajectory for radiopaque landmark-based measurements. At this point, the patient developed decreased end-tidal carbon dioxide ($ETCO_2$) and oxygen saturation, with significant ductal spasm and reduced effective pulmonary flow. Continuous adrenaline infusion was started. The BHW guidewire was replaced with an exchange-length 0.014″ wire. With guidewire support, an Inspiron 4 × 19 mm stent was implanted in the ductus arteriosus (Figure 4).

The patient showed transient improvement in saturation and $ETCO_2$, but a few minutes later developed significant hypoxemia, decreased $ETCO_2$, and bradycardia. Acute stent thrombosis was identified, and in situ alteplase was administered as a 0.05 mg/kg bolus, followed by continuous infusion at 0.5 mg/kg/h. Sequential balloon angioplasty of the entire stent with a Trek 4 × 12 mm balloon catheter was performed. These measures restored

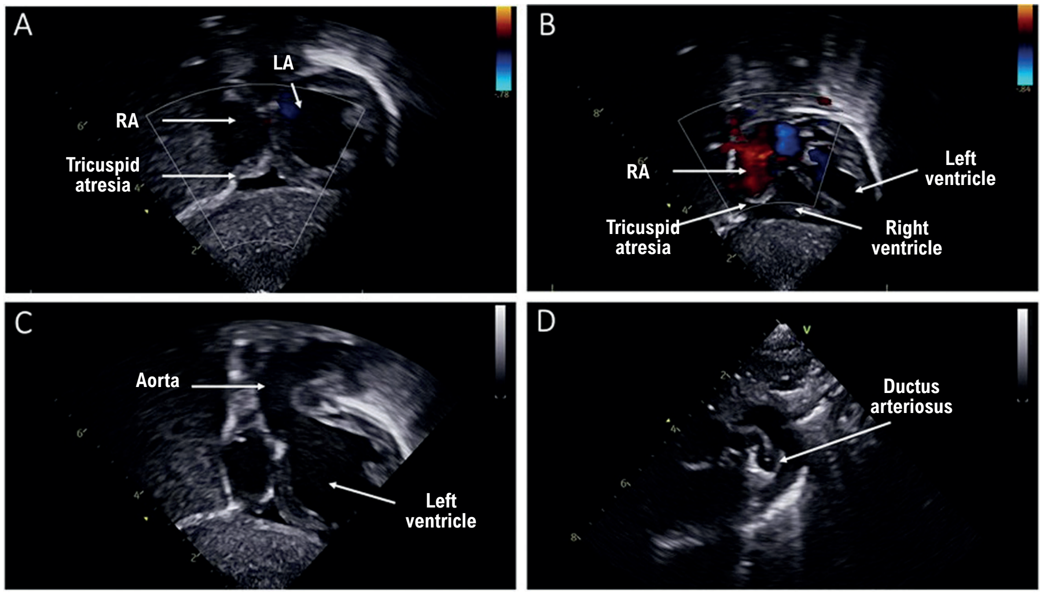

Figure 1 – *Transthoracic echocardiography. A and B) Patent foramen ovale and atretic tricuspid valve. C) Left ventricle connected to the aorta. D) Tortuous patent ductus arteriosus. LA: left atrium; RA: right atrium.*

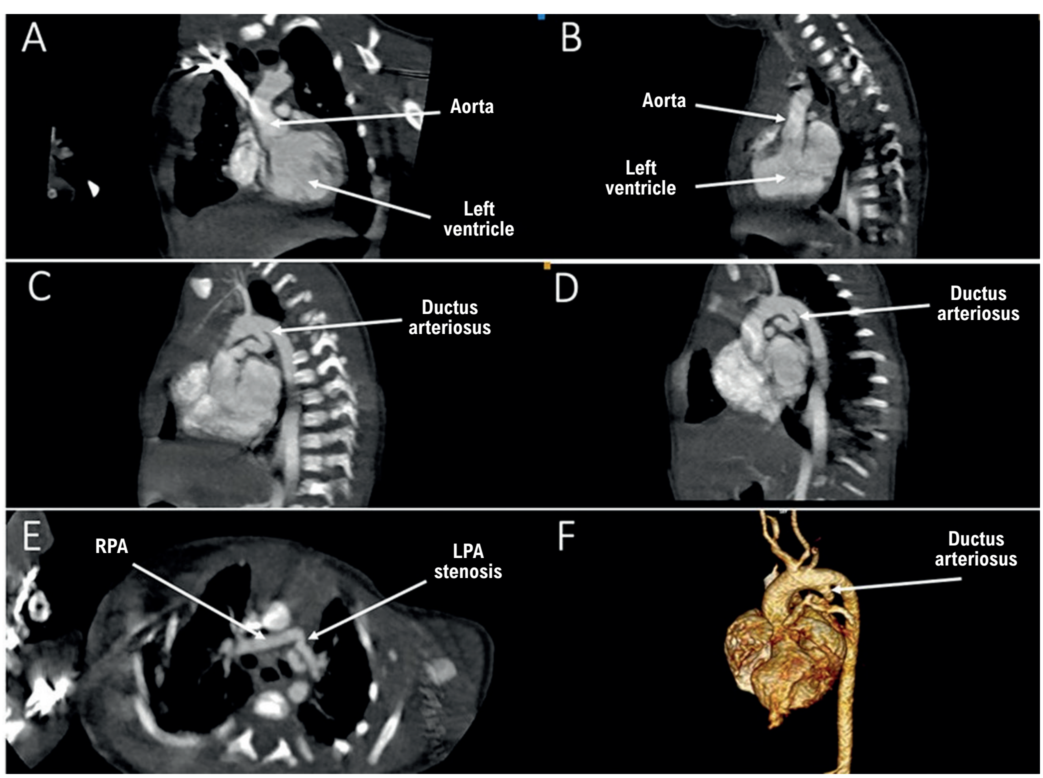

Figure 2 – *Aortic computed tomography angiography. A and B) Left ventricle connected to the aorta. C and D) Markedly tortuous patent ductus arteriosus. E) Stenosis at the origin of the left pulmonary artery. E) Three-dimensional reconstruction showing the anatomy of the ductus arteriosus. LPA: left pulmonary artery; RPA: right pulmonary artery.*

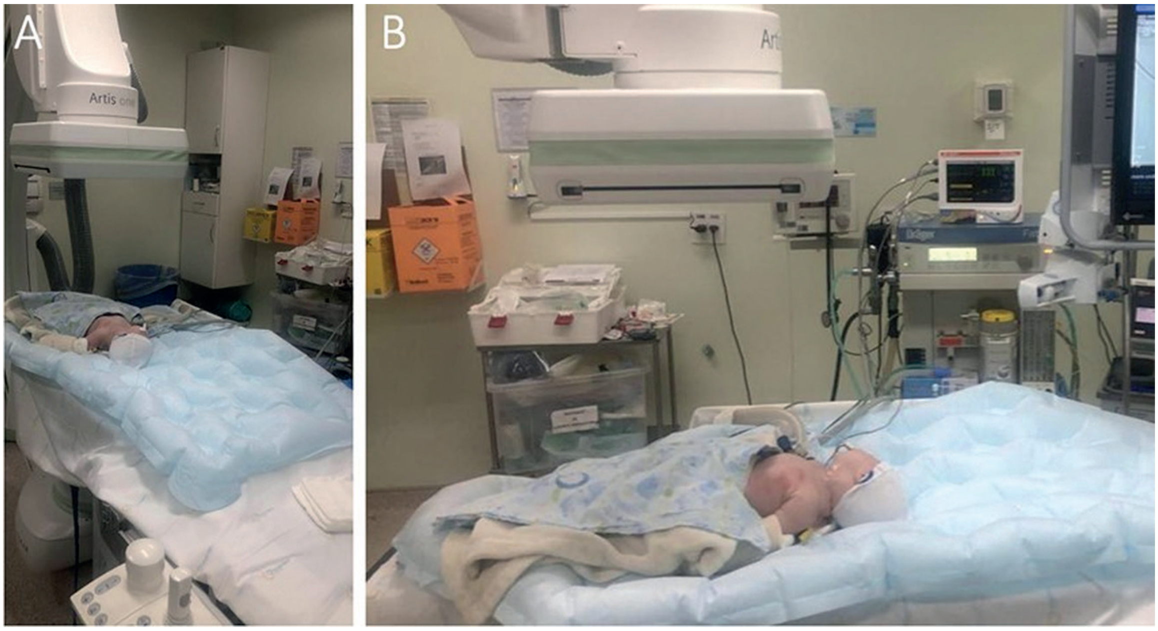

Figure 3 – *A and B) Image showing the patient positioned in reverse orientation on the catheterization table (with the cranial portion away from the radiation source).*

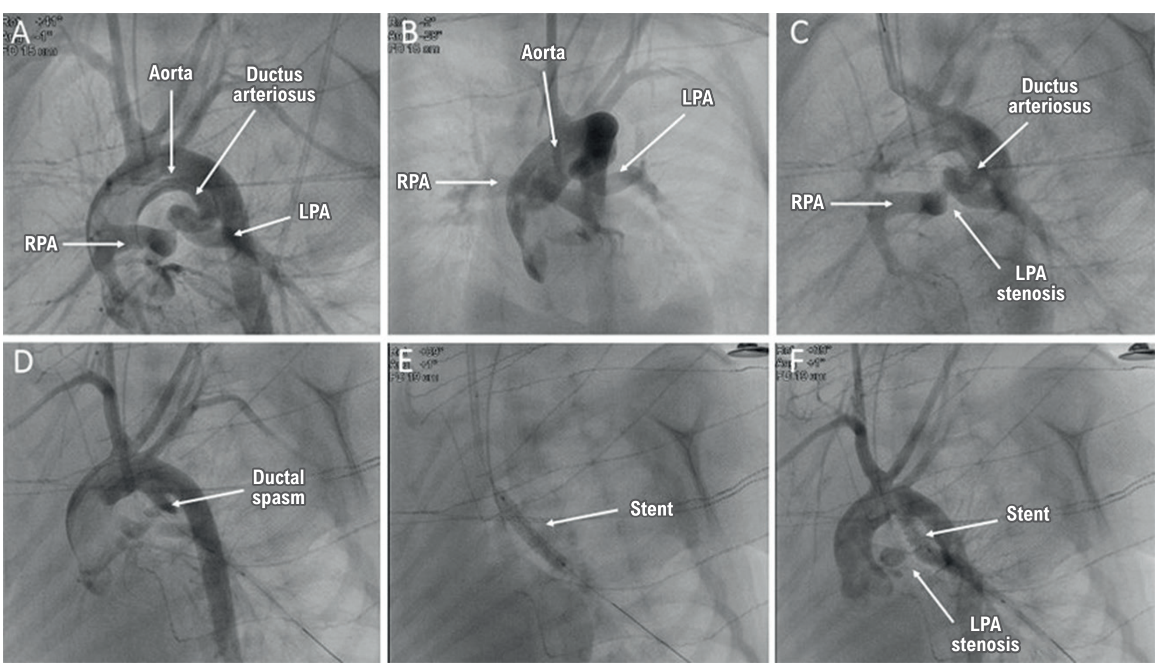

Figure 4 – *Percutaneous intervention. A, B, and C) Angiograms obtained via carotid access showing a tortuous ductus arteriosus and stenosis at the origin of the left pulmonary artery. D) With the support of a 0.014" BHW guidewire positioned in the left pulmonary artery, a Mini Trek 2 × 8 mm balloon catheter was placed along the ductal trajectory; at this moment, ductal spasm was observed. E) An Inspiron 4 × 19 mm stent was implanted in the ductus arteriosus. F) Angiogram showing preserved flow through the ductal stent but with worsening stenosis at the origin of the left pulmonary artery. LPA: left pulmonary artery; RPA: right pulmonary artery.*

ductal flow and partially improved heart rate, oxygen saturation, and $ETCO_2$.

Stenosis at the origin of the left pulmonary artery significantly restricted flow to the pulmonary trunk and right pulmonary artery. Proximal optimization technique (POT) was used with a 4 × 8 mm balloon (inflated to burst pressure). With the support of a JR 4F catheter, a 0.014″ guidewire was placed in the right pulmonary artery through the lateral mesh of the prior stent. Balloon angioplasty of the right pulmonary artery origin was then performed with a Trek 3 × 12 mm balloon (opening the lateral mesh of the previous stent) (Figure 5). After balloon angioplasty, the guidewire was withdrawn from the right pulmonary artery, but the stenosis at the origin of the left pulmonary artery persisted.

The 0.014″ guidewire was then repositioned in the right pulmonary artery, and an Inspiron 3.5 × 9 mm stent was implanted through the lateral mesh of the previous stent, directed toward the origin of the left pulmonary artery and the pulmonary trunk (Figure 6). The introducer was removed, and manual hemostatic compression and compressive occlusive dressing were applied.

The patient was transferred to the intensive care unit on mechanical ventilation and continuous intravenous adrenaline (0.1 mcg/kg/min). Alteplase continued for another 3 hours but discontinued due to bleeding at the puncture and central venous access sites. Grade I left intracranial hemorrhage was detected on post-procedure transfontanellar ultrasound, which normalized on follow-up 3 months later. The patient had infectious complications such as pneumonia and sepsis with positive blood cultures for *Pseudomonas aeruginosa*, treated with broad-spectrum antibiotics. He also experienced extubation failure and prolonged mechanical ventilation (total of 49 days). Hospitalization was prolonged, with discharge 3 months after the procedure. Transthoracic echocardiography showed patent stents with adequate flow through the ductus arteriosus and pulmonary branches. Clinically, the patient remains hemodynamically stable, with oxygen saturation of 84% in room air, and is being followed for planned Glenn surgery.

Discussion

Tricuspid atresia with pulmonary atresia is one of the most complex challenges in interventional pediatric cardiology, especially because of the absolute dependence on ductal flow for pulmonary perfusion in the neonatal period.[4,5] This case illustrates the multiple layers of anatomic and technical complexity involved in the management of these patients, highlighting both the therapeutic possibilities and the inherent limitations and complications of interventional procedures in high-risk neonates.

Ductal stenting has emerged as a viable and often preferable alternative to surgical shunts in selected neonates,

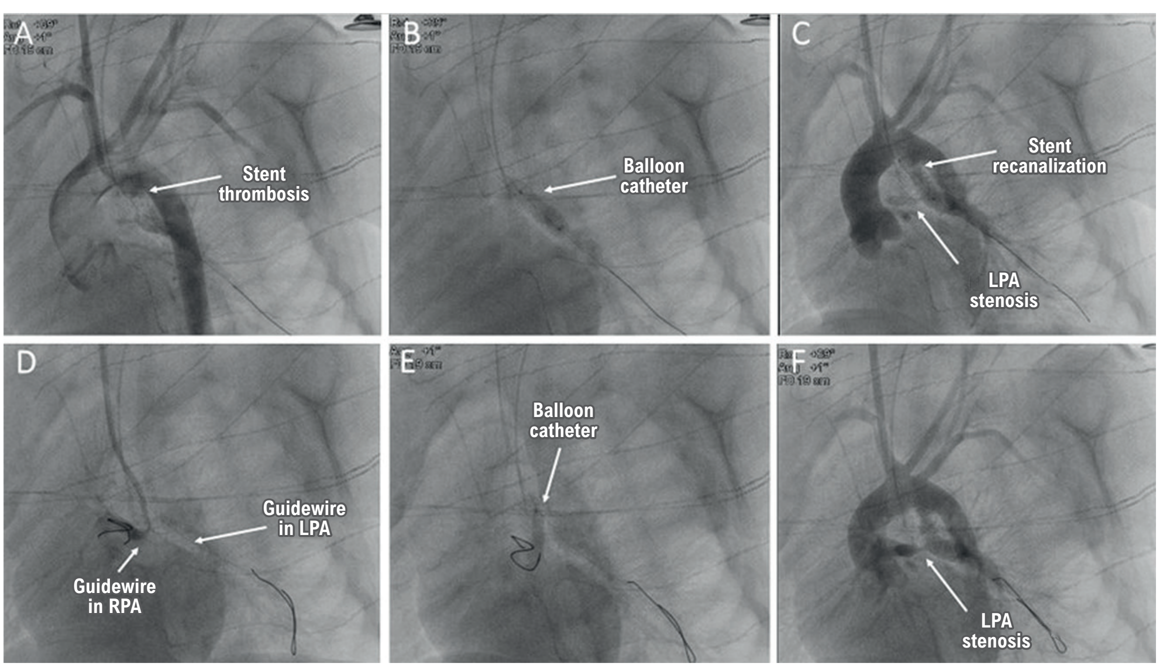

Figure 5 – *Percutaneous intervention. A) Angiography showing acute thrombosis of the ductal stent. B) Angioplasty with a 4 × 12 mm Trek balloon catheter along the entire length of the stent. C) Angiography demonstrating effective recanalization of the stent. D) Guidewire positioned in the right pulmonary artery through the lateral struts of the stent. E) Angioplasty with a 3 × 12 mm Trek balloon catheter (opening the lateral struts of the previous stent). F) Guidewire withdrawn from the right pulmonary artery, showing persistence of stenosis at the origin of the left pulmonary artery. RPA: right pulmonary artery; LPA: left pulmonary artery.*

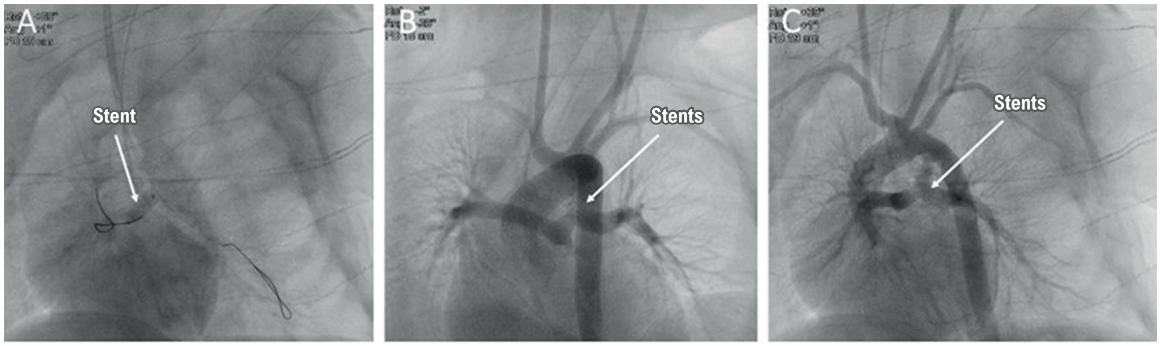

Figure 6 – *Percutaneous intervention. A) Implantation of an Inspiron 3.5 × 9 mm stent through the lateral mesh of the previous stent. B and C) Angiograms showing adequate flow through the stent assembly from the ductus arteriosus to both pulmonary arteries.*

offering significant advantages in terms of perioperative morbidity and mortality.[6,7] Recent comparative studies have shown similar survival rates between the two therapeutic modalities, but with a lower incidence of immediate complications in the percutaneous intervention group.[8,9] However, appropriate patient selection remains critical, considering factors such as neonatal weight, ductal anatomy, associated pulmonary stenoses, and pre-procedure hemodynamic stability.[10,11]

The angle formed between the aortic end of the ductus arteriosus and the aortic arch, as seen in this case, is an anatomical limitation that requires significant technical adaptations, including changes in patient positioning and careful selection of the vascular access route.[12-14] The extreme ductal tortuosity[15] combined with the short length of the pulmonary branches in a newborn complicates coronary guidewire navigation and the support it provides for stent delivery. The use of specific materials such as a microcatheter and more than one 0.014″ guidewire positioned simultaneously makes the procedure feasible. Pre-existing pulmonary branch stenosis prior to stent implantation is a risk factor for worsening stenosis and total occlusion of a pulmonary branch.[16] Techniques such as positioning a 0.014″ guidewire in each pulmonary branch during stent implantation can increase procedural safety by maintaining patency of the stenotic pulmonary branch and facilitating the opening of the stent's lateral mesh. In this case, this technique was not possible because the patient developed acute clinical deterioration during guidewire manipulation within the ductus, causing ductal spasm.

The occurrence of acute stent thrombosis is one of the most feared complications in neonatal procedures, with an incidence of 2% to 3%. Predisposing factors include elevated hematocrit, prolonged procedure time, endothelial trauma during manipulation, and activation of the coagulation cascade secondary to the prosthetic material. Immediate treatment with local fibrinolytics, as demonstrated in this case with alteplase, has been effective, allowing successful recanalization with low risk of systemic bleeding. Sequential mechanical angioplasty of the stent also restores adequate flow quickly and sustainably.[10,17]

The technique of stent implantation through the lateral mesh of a previously placed stent ("stent-in-stent") used to address stenosis at the origin of the left pulmonary artery represents a significant technical innovation in pediatric interventional cardiology. The literature documents growing experience with this technique in various anatomical situations, demonstrating medium-term safety and efficacy.[18,19]

The complications observed in this case, including grade I intracranial hemorrhage and hospital-acquired infections, reflect the fragility of neonates undergoing complex and prolonged interventional procedures. Intracranial hemorrhage, possibly related to the use of fibrinolytics, is a known but relatively rare complication, requiring strict neurological monitoring in the post-procedural period.[20,21] The development of nosocomial infections in critically ill patients on prolonged mechanical ventilation is an additional challenge in post-intervention management, demanding rigorous prevention and early treatment protocols. The favorable medium-term outcome, with maintained stent patency and hemodynamic stability, supports the efficacy of the chosen therapeutic strategy and the use of this approach as an effective bridge to subsequent surgical staging in selected patients.[22,23]

Conclusion

This case demonstrates the feasibility and efficacy of hemodynamic intervention in neonates with type 1A tricuspid atresia. Ductal stenting allowed for adequate pulmonary blood flow and clinical stabilization. The observed complications, including acute stent thrombosis, worsening of left pulmonary artery origin stenosis after ductal stenting, and hemodynamic instability, reflect the complexity of neonatal procedures and the need for rapid and effective medical (fibrinolytic) and interventional (balloon angioplasty of the thrombosed stent, POT technique, balloon and stent angioplasty through the lateral stent mesh) strategies. The favorable medium-term outcome, with preserved pulmonary flow and ventricular function, supports the use of this technique as a bridge to future surgical staging in selected patients.

Author Contributions

Conception and design of the research and writing of the manuscript: Lombardi JG, Gardenghi G; acquisition of data: Lombardi JG, Calamita PC, Barreto MRP, Ficht LSN, Gardenghi G; analysis and interpretation of the data: Lombardi JG, Calamita PC, Barreto MRP; critical revision of the manuscript for intellectual content: Calamita PC, Barreto MRP, Ficht LSN, Gardenghi G.

Potential Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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Study Association

This study is not associated with any thesis or dissertation work.

Ethics Approval and Consent to Participate

This study was approved by the Ethics Committee of the CEP do Hospital de Urgëncias de Goiás under the protocol number 85497418.2.0000.0033. All the procedures in this study were in accordance with the 1975 Helsinki Declaration, updated in 2013. Informed consent was obtained from all participants included in the study.

Use of Artificial Intelligence

The authors did not use any artificial intelligence tools in the development of this work.

Availability of Research Data

The data cannot be made publicly available due to legal considerations related to Brazil's General Data Protection Law, as the data could allow the identification of the patient in the case in question.

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