

# Primary Ventricular Palliation in Hypoplastic Left Heart Syndrome: A Case Report

Giovana Ferreira Vaz <sup>1,\*</sup>, Marcos Barbosa Guimarães Carvalho <sup>1</sup>, Maria Eduarda Santos Gomes <sup>1</sup>, João Vitor Mendes da Silveira <sup>1</sup>, Leonardo Sardinha de Paula <sup>1</sup>, Mirna de Sousa <sup>2</sup>, Viviane Soares <sup>1</sup>

<sup>1</sup> Evangelical University of Goiás (UniEVANGÉLICA), Anápolis, Goiás, Brazil.

<sup>2</sup> Governor Otávio Lage State Emergency Hospital (HUGOL), Goiânia, Goiás, Brazil.

\* Correspondence: giovanafv03@gmail.com.

**Abstract:** Hypoplastic Left Heart Syndrome (HLHS) is a complex congenital heart disease significantly contributing to neonatal mortality. It is characterized by the severe underdevelopment of left-sided heart structures, obstructing systemic circulation and necessitating a patent ductus arteriosus and atrial communication for survival. This case involves a 35-year-old hypertensive and diabetic mother who received prenatal care through the Unified Health System (SUS). HLHS was identified via two fetal echocardiograms during pregnancy. Delivery occurred by emergency cesarean section following 50% premature placental abruption. The neonate presented an Apgar score of 8/8, weighing 2080g, with a length of 46cm and head circumference of 31.5cm. Management began on April 20, 2023, through hybrid surgery, including a 7x24 mm ductal stent, bilateral pulmonary artery banding, and a pulmonary trunk banding. Notably, this was the first successful procedure of its kind in Goiás. Postoperatively, the patient overcame *Acinetobacter*-induced sepsis diagnosed on April 22, 2023. On September 10, hospitalization was required for nutritional failure, successfully managed with a nasogastric tube. The patient is currently undergoing clinical follow-up and monitoring for sequelae following the second stage of surgical palliation. This case underscores the critical role of early diagnosis and advanced surgical interventions in improving neonatal survival and long-term outcomes.

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## 1. Introduction

Hypoplastic Left Heart Syndrome (HLHS) can be defined as a set of congenital cardiovascular malformations related to the underdevelopment of the left side of the heart. This syndrome involves atresia, stenosis, or hypoplasia of the aortic and/or mitral valves and hypoplasia of the ascending aorta and the aortic arch. These underdeveloped structures cannot support the blood flow necessary for systemic circulation and, thus, surgical correction or cardiac transplantation becomes necessary [1]. The incidence of HLHS is 0.2 – 0.4 per 1,000 live births and, when untreated, it is responsible for 25% of deaths related to congenital heart diseases that occur in the first weeks of life [2].

Most cases of HLHS are diagnosed via prenatal ultrasonography, allowing for parental counseling, appropriate delivery planning, and the rapid initiation of treatment at birth. After birth, the diagnosis must be confirmed by performing an echocardiogram,



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which aims to identify the specific anatomical aspects of the case and its hemodynamic consequences [3].

The clinical presentation depends on hemodynamic factors such as restriction of blood flow at the atrial level (which varies according to the presence of an atrial septal communication), persistence of the ductus arteriosus, and pulmonary vascular resistance. In cases where the restrictive atrial septal communication is absent or small, there is a significant increase in pressure in the left atrium that is transmitted retrogradely to the pulmonary circulation, leading to the development of severe left atrial hypertension and subsequent pulmonary venous congestion. Consequently, symptoms of respiratory distress and low systemic output appear immediately or shortly after birth [4].

Persistent atrial septal communication in newborns is considered a protective factor for HLHS, making the presentation of symptoms more progressive and later, rather than abrupt. However, these neonates also require the presence of the ductus arteriosus to allow systemic blood flow and coronary circulation [2,5]. In addition to ductal patency, it is also necessary for the flow through the ductus to be in the pulmonary-to-aortic direction, which occurs due to the high pulmonary vascular resistance characteristic of the fetal pattern still present in newborns. However, at some point, this ductus begins to close and pulmonary resistance starts to decrease, thus initiating the symptoms. Initially, arterial oxygen saturation (SatO<sub>2</sub>) is increased; subsequently, there may be pulmonary edema, decreased SatO<sub>2</sub>, tachypnea, and respiratory failure [4,6,7]. Concomitantly, there is poor feeding, progressing to lethargy, pallor, cold extremities, weak pulses, delayed capillary refill, hepatomegaly, shock, facial edema, and a full fontanelle [2,4].

Immediate clinical treatment consists of intravenous prostaglandin infusion to maintain the ductus arteriosus patent and life support with mechanical ventilation and vasoactive amines [8]. Surgical treatment is carried out through a three-stage surgery with palliative procedures, commonly identified as staged Fontan palliation, since an effectively corrective treatment is not possible. For the first stage, which occurs in the first weeks of life, there are three surgical options: the classic Norwood procedure, the Norwood procedure with Sano modification, and the hybrid procedure [1, 9].

The classic Norwood procedure, with the Blalock-Taussig shunt, consists of creating a neo-aorta by connecting the hypoplastic aorta to the pulmonary trunk, ensuring systemic and coronary blood flow. In the classic procedure, pulmonary blood flow is ensured by creating a "shunt" between the subclavian artery and the ipsilateral pulmonary artery via a synthetic tube. In the Sano variant, this flow is ensured through the grafting of a tube from the right ventricle to the pulmonary trunk [10,11].

The hybrid procedure is performed by taking advantage of the natural physiology of the fetal circulation characteristic of the syndrome. In this procedure, a stent is placed in the patent ductus arteriosus to keep it open, thus ensuring systemic and coronary blood flow. Banding of the pulmonary artery branches is also performed, creating branch stenosis and maintaining high pressure in the pulmonary trunk so that flow through the stent remains in the pulmonary-to-aortic direction [10]. In all procedures, the atrial septal communication must be maintained to ensure both systemic and pulmonary blood flow [12]. The hybrid procedure offers advantages such as the ability to more safely treat low-birth-weight children and is associated with lower morbidity and mortality. Thus, it constitutes an alternative for treating patients with HLHS in centers where mortality is high for Norwood surgery [12].

The first procedure, whether Norwood or hybrid, must be performed in the first days of life and must be replaced shortly thereafter, as children's growth is rapid and the grafted tubes do not keep pace with this growth. Consequently, the second surgery, known as Glenn palliation, should be performed between 4 and 9 months of age. Glenn surgery involves anastomosing the superior vena cava to the pulmonary territory. At this age, the demand for blood flow and venous return in children is greater from the upper part of the body, which is sufficient to maintain adequate oxygen saturation.

The third and final stage of the palliative process, the Fontan procedure itself, is performed by connecting the inferior vena cava to the pulmonary artery [4]. After this procedure the child ceases to exhibit symptoms of hypoxia, as the procedure prevents the mixing of oxygenated and deoxygenated blood in the atria, ensuring that oxygenated blood stays inside the heart and deoxygenated blood remains outside of it [2].

This case report on HLHS is significant due to the severity of this heart disease, which has a major impact on infant mortality; its early diagnosis and appropriate treatment have the potential to change this scenario. Furthermore, the case in question refers to the first hybrid procedure successfully performed by the SUS (Unified Health System) in the State of Goiás. Despite the sequelae currently under treatment, the result is surprising and promising for the care of children with this type of diagnosis, as the chances of success for Fontan Palliation procedures remain low. The procedure was performed at a public hospital maintained by the Unified Health System (SUS), which provides care across multiple specialties beyond cardiovascular services. It is the only center featuring a specialized team for this high-cost intervention, which requires a hybrid operating room equipped with high-resolution diagnostic tools for hemodynamic assessment (e.g., outflow gradient across the pulmonary band) and real-time imaging guidance. Thus, the objective of the study was to report the case of hypoplastic left heart syndrome in which the first palliative surgery performed in the state of Goiás was successful.

## 2. Case Report

The newborn patient is male, the son of a hypertensive and diabetic mother with no other reported conditions. Delivery was by cesarean section, indicated due to premature placental abruption. The newborn was received in the delivery room with an axillary temperature of 36.5 °C on April 14, 2023, at 05:14. After birth, upper airway and gastric suctioning were performed, along with external stimulation, use of a manual resuscitator with positive pressure, and 21% oxygen supply. The procedures were performed because the newborn did not breathe or cry at birth. The cord was clamped 1 minute before, skin-to-skin contact with the mother was provided, and the Apgar score was 8/8. Anthropometric measurements at birth were weight equal to 2.080 kg, height of 46 cm, and head circumference of 31.5 cm.

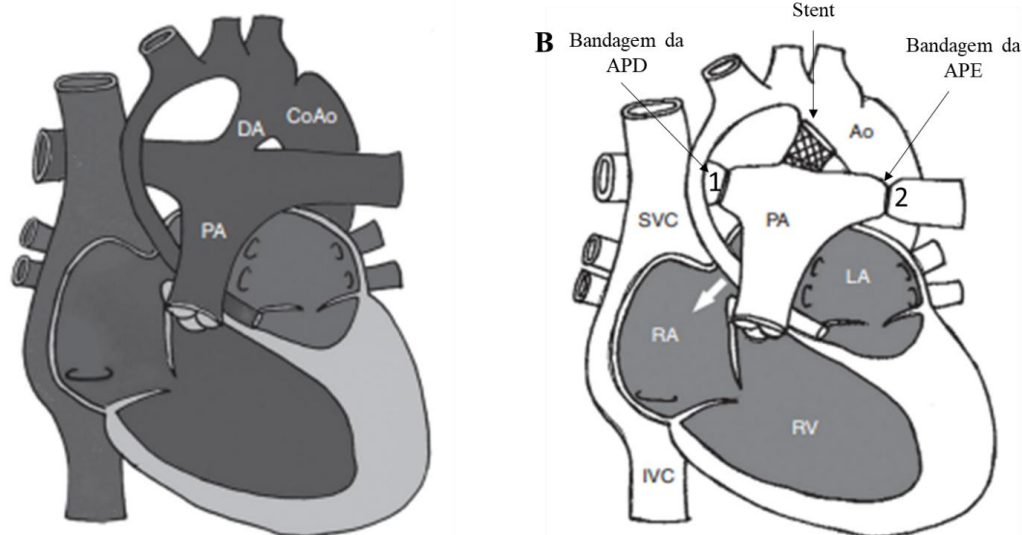
He was characterized as preterm, with a gestational age of 35 weeks and 5 days based on an ultrasound (USG) performed in the first trimester, and the presence of a single umbilical artery was identified. He showed no apparent morphological alterations or lesions on examination. A further limitation of this study is the absence of karyotyping, array-CGH, or screening for associated genetic syndromes (e.g., Turner syndrome, Trisomy 18, or GJA1 variants). The patient's phenotype was not suggestive of any classical syndrome. Genetic evaluation is not a routine practice in these services due to structural and resource limitations.

The newborn was previously diagnosed with cardiac alterations during a routine USG at 22 weeks and 3 days of gestation, with changes in the structure of the chambers being found. Following the diagnosis, two fetal echocardiograms were documented in medical records, both concluding with HLHS. However, the study faces a documentation limitation, as the original fetal echocardiogram reports were unavailable for presentation. Nevertheless, a neonatal echocardiogram was performed on April 15, 2023, and reported the following day, confirmed HLHS. The findings included a univentricular heart due to the absence of the left atrioventricular connection, a non-restrictive patent foramen ovale (PFO), and left ventricular hypoplasia. The left ventricle remained hypoplastic with a virtual cavity across all serial examinations.

The mitral and aortic valves were hypoplastic (aortic annulus: 2.34 mm), exhibiting minimal anterograde flow. The ascending aorta measured 4.3 mm, the sinotubular junction 2.7 mm, and the transverse arch 4.4 mm, with the latter showing reverse flow. The right-sided chambers showed moderate-to-severe dilatation, and the right ventricle was

hypertrophied, functioning as a single systemic ventricle. The pulmonary trunk was dilated, and the right and left pulmonary arteries were confluent. The ductus arteriosus was patent and wide (pulmonary end: 5.7 mm; aortic end: 6.4 mm; length: 7.9 mm), with bidirectional flow predominantly from the pulmonary trunk to the ascending aorta. An intact interventricular septum was confirmed by both computed tomography angiography (CTA) and all serial echocardiograms. Six days after birth, the newborn underwent the first stage of the Fontan palliation treatment, and the procedure of choice was the hybrid (Figure 1).

**Figure 1.** A. Illustration of a heart with Hypoplastic Left Heart Syndrome. At birth, children with HLHS depend on blood pumped by the right ventricle into the pulmonary artery (PA) to provide blood to the systemic circulation and end organs via a patent ductus arteriosus (DA). In many cases, coarctation of the aorta (CoAo) is also present. B. Schematic drawing of the hybrid procedure showing RPA and LPA banding to restrict pulmonary blood flow and the placement of a stent in the ductus arteriosus to maintain patency. RPA: right pulmonary artery; LPA: left pulmonary artery; SVC: superior vena cava; IVC: inferior vena cava; PA: pulmonary artery; Ao: aorta; LA: left atrium; RA: right atrium. Source: Adapted from Yabrodi and Mastropietro [13].

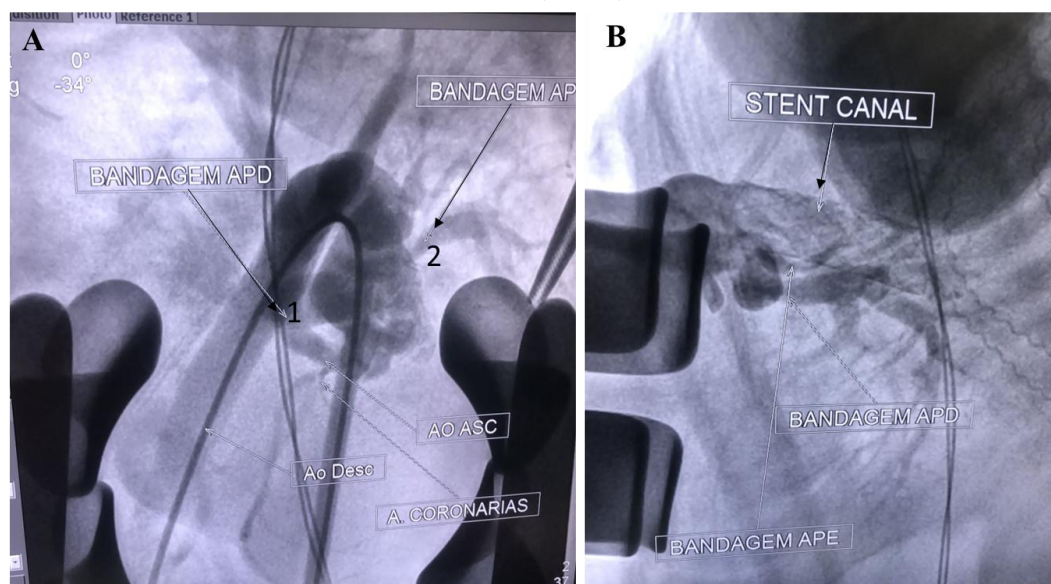


During the surgery, a 7x24mm stent was placed in the ductus arteriosus via cardiac catheterization, and pulmonary artery banding (Figure 2) were performed, with the banding on the left side being placed more tightly. In this neonate, the ductus arteriosus measured 6.3 mm at the pulmonary end and 5.8 mm at the aortic end (length: 22.1 mm), justifying the selection of a 7x24 mm stent. This allowed for expansion to a 7.1 mm diameter over a length of 23.7 mm, ensuring the maintenance of ductus-dependent systemic flow. This strategy follows the standard hybrid protocol for high-risk HLHS.

The stent was inflated to a pressure of 10 atm to ensure that the manufacturer-specified nominal diameter was achieved. Pressures below this level may result in under-expansion, leading to residual gradients, an increased risk of thrombosis, stent displacement, or embolization. Conversely, pressures exceeding the recommendations may cause over-dilation, potentially leading to stent deformation or traumatic injury to the vessel/tract [14]. Pulmonary artery banding is a technique used in pediatric cardiac surgeries that consists of encircling the artery with a band to reduce its caliber at that site, inducing stenosis in the pulmonary branches with the aim of reducing downstream blood flow.

In this hybrid procedure, the main objective was to increase the pressure in the pulmonary trunk (upstream) to ensure the maintenance of flow through the stent from the pulmonary trunk to the aorta, making it possible to maintain systemic and coronary flow. This flow can be elevated in certain heart conditions, such as HLHS, and can cause hypertrophy of the pulmonary vasculature or pulmonary hypertension if not corrected or adjusted. Hybrid palliation is a combination of surgery and catheterization and, in this report, took place in a hybrid room involving both a surgical team and a hemodynamics team. The procedure lasted 4 hours and consisted first of the pulmonary artery banding (surgical part) followed by the ductal stent implantation (hemodynamic part). The criterion for choosing the hybrid procedure over the Norwood procedure in this case was the high mortality rate of the Norwood procedure at the hospital service where the case was treated. Following a global trend, in centers where Norwood outcomes are not favorable, the hybrid procedure is pursued, as it has lower morbidity and mortality.

**Figure 2.** A. Catheterization performed during the procedure: placement of right pulmonary artery (RPA) (arrow 1) and left pulmonary artery (LPA) (arrow 2) banding. B. Location of the stent within the ductus arteriosus (arrow).



Following the surgery (after two days), the newborn developed sepsis with an identified etiological agent (*Acinetobacter*), which was managed with Meropenem (120 mg/kg/day), Vancomycin (40 mg/kg/day), and Fluconazole (6 mg/kg/day). Following hospital discharge, the infant remained on nutritional support via a nasoenteral tube (Infatrini 400); however, weight gain between August and September was poor, increasing from 3,915g to only 4,130g. In September, the patient was readmitted due to failure to thrive and diarrhea. The condition was resolved with the administration of multivitamins (0.5 ml once daily), zinc (2.5 ml), and an elemental formula diet (Neocate) at 70 ml every 3 hours via both nasoenteral and oral routes.

Postoperative data from the hybrid procedure indicated atrial communication that was difficult to assess, with pressure gradients from the pulmonary trunk to the right pulmonary artery of 49 mmHg and to the left pulmonary artery of 72 mmHg. There was no available data regarding the retrograde gradient, and mild-to-moderate right ventricular dysfunction was noted. Echocardiograms performed on August 30, 2023, and September 29, 2023, documented pulmonary trunk-left pulmonary artery gradients of 86 and 81 mmHg, respectively, with the banding classified as tight. An increasing retrograde aortic gradient (27 and 23 mmHg) indicated progressive compromise of coronary perfusion.

These findings, combined with poor weight gain and ductal stent stability, supported the decision to proceed with the Norwood-Glenn procedure in November 2023.

The second stage of correction is not yet offered at the service where the first procedure was performed. For this reason, the patient was transferred to the Heart Institute of the Faculty of Medicine of the University of São Paulo in São Paulo (InCor FMUSP) to undergo the second stage of palliation, the Glenn surgery. This stage was performed in São Paulo at InCor and, after the procedure, the patient was in a serious general state, requiring the use of ECMO (extracorporeal membrane oxygenation). Currently, the patient has been weaned from ECMO and is undergoing adjustments and monitoring for postoperative sequelae. The Norwood-Glenn procedure was performed on November 21, 2023, when the infant was 7 months and 7 days old. However, there is a documentation limitation regarding the lack of records for the patient's weight at the time of the procedure. The last documented weight prior to transfer to InCor was 3.240 kg on June 29, 2023.

### 3. Discussion

HLHS is a severe condition where those affected are typically born in term and initially appear healthy; however, after the onset of the circulatory transition, with the drop in pulmonary vascular resistance and closure of the ductus arteriosus, systemic perfusion decreases and symptoms of low systemic output, such as hypoxemia, acidosis, and shock, appeared [2]. In the reported case, due to the prenatal diagnosis, it was possible to plan the delivery and prepare the obstetrics and neonatology teams to receive the patient, initiate clinical treatment, and provide rapid referral for surgical treatment. Only one of these conditions was observed, since, although the patient was born well with a 1-minute Apgar score of 8, the birth was premature. However, the early delivery date may be related to the mother's obstetric data, such as advanced maternal age, type 2 diabetes mellitus, and high blood pressure.

Prenatal diagnosis was achieved through ultrasonography. It is easily identified by the presence of a small left ventricular cavity in the fetal four-chamber view. There are also the absence of a common atrioventricular junction and no ventricular septal defect. The ascending aorta is typically small and exhibits retrograde or bidirectional flow in the presence of aortic stenosis [3]. In addition to the ultrasound diagnosis, acute maternal hyperoxygenation tests can be performed, which are responsible for increasing pulmonary blood flow by decreasing pulmonary vascular resistance. In patients with HLHS, the examination may reveal reduced vasodilatory capacity of the lung [3]. The diagnosis of the patient in this study was made through these two processes, with fetal ultrasounds indicating cardiac alterations and, subsequently, HLHS was identified through a fetal echocardiogram (ECHO) fetal.

Generally, cardiac murmurs are absent; if present, they are non-specific. The second heart sound is loud and single, and hepatomegaly may occur due to hypoxia [15]. However, dynamic instability promotes the emergence of early signs and symptoms, such as congestive state, low systemic output, pallor, dyspnea, discomfort, irritability, and variable cyanosis [16]. It is a syndrome with a low incidence, yet it is responsible for a large portion of deaths among newborns with congenital heart diseases. It is preferable that this syndrome be diagnosed during prenatal care to provide treatments and measures in a rapid, objective, and correct manner postpartum [2].

An option for the first stage of palliation is the hybrid procedure. In the case reported, the patient underwent the hybrid procedure, which involved placing a 7x24mm stent in the ductus arteriosus and performing pulmonary artery banding, with the banding on the left side being more tightly applied. This procedure consisted of bilateral banding of the pulmonary artery branches and placement of a stent in the ductus arteriosus, without cardiopulmonary bypass [10]. This process is less invasive; however, it requires closer follow-up, more frequent interventions, and a shorter interval between stages I and II of the correction [4,17]. As indicated in studies, even though it is a less invasive procedure, it must

be closely monitored, as seen in the patient who developed sepsis and subsequently diarrhea and dehydration [18,19,20]. Additionally, difficulty in gaining weight and adapting to an oral diet was observed.

The hybrid palliation performed on the newborn was a viable alternative to replace the classic Norwood procedure, especially in severe cases or in patients with borderline left ventricles [12]. As the survival rates of children with HLHS have increased, concern has shifted toward the long-term survival and quality of life of affected individuals. Studies have been conducted with this objective, using various tests to evaluate neurodevelopment and quality of life. Such parameters were assessed in studies through behavioral tests [4]. Other studies have demonstrated outcomes such as heart transplant listing, hospitalization for heart failure, sustained ventricular tachycardia, protein-losing enteropathy, or thromboembolic events as consequences in patients who have undergone Fontan palliation [15].

#### 4. Conclusion

The patient in the present case has Hypoplastic Left Heart Syndrome (HLHS), diagnosed during prenatal exams (ultrasonography and echocardiogram). He was born relatively well but progressed to present typical symptoms of the syndrome. Six days after birth, the initial Fontan palliation procedure was performed; the chosen approach in this case was the hybrid procedure. This surgery was the first of its kind to be successfully performed by the SUS (Unified Health System) in the State of Goiás. The patient progressed well postoperatively, was monitored, and subsequently underwent the second stage at InCor-FMUSP. He was in critical condition but is currently being followed up for postoperative sequelae. Thus, survival and success in hybrid corrections depend on effective prenatal monitoring with exams available for early detection.

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**Research Ethics Committee Approval:** This case study was submitted to and approved by the Research Ethics Committee (CEP) of Evangelical University of Goiás under number CAAE: 75804023.9.0000.5076, with the patient's consent obtained through the signature of the Free and Informed Consent Form (TCLE), following Resolution 466/2012 of the National Health Council (CNS), which governs research involving human beings, and Circular Letter 166/2018, which governs case reports.

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**Conflicts of Interest:** All other authors declare no conflicts of interest.

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