

## PEDIATRIC CARDIOLOGY

# Patterns of Anomalous Pulmonary Venous Connection/Drainage in Hypoplastic Left Heart Syndrome: Diagnostic Role of Doppler Color Flow Mapping and Surgical Implications

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Differentiation between anomalous connection and anomalous drainage of the pulmonary veins in hypoplastic left heart syndrome is important before either the Norwood procedure or heart transplantation is performed. To determine the prevalence of echocardiographically detected anomalous connection or drainage, or both, of pulmonary veins in patients with this syndrome, preoperative two-dimensional echocardiographic and Doppler color flow mapping studies of 317 patients who underwent the stage I Norwood procedure were reviewed. The term "connection" was used to describe the precise anatomic attachment of the pulmonary veins and the term "drainage" to describe the physiologic end point of pulmonary venous flow.

Twenty patients (6.3%) had anomalous connection or drainage, or both, of the pulmonary veins by preoperative echocardiographic and Doppler examination. The subcostal and suprasternal

scans best showed the anatomic details of the pulmonary veins. All these patterns were confirmed intraoperatively and could be grouped as follows: 1) partial anomalous connection and drainage (two patients); 2) total anomalous connection and drainage (eight patients); 3) normal connection with total anomalous drainage (eight patients); and 4) normal connection with partial anomalous drainage (two patients). The advantage of adding Doppler color flow mapping to two-dimensional echocardiography and conventional Doppler study was clearly demonstrated in the detection of small accessory vertical veins, their course and the presence or absence of obstruction. Doppler color flow mapping was especially helpful in detecting anomalous drainage of the right pulmonary veins to the right of the superior attachment of the septum primum.

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Precise definition of the connection sites of the pulmonary veins in hypoplastic left heart syndrome is important before either heart transplantation or reconstructive surgery (Norwood procedure followed by subsequent Fontan procedure) is performed. A false diagnosis may result in prolongation of the surgical procedure as the surgeon tries to define precisely the connection and drainage pattern of the pulmonary veins. Although there may be no physiologic hazard in not repairing an unobstructed total anomalous pulmonary venous connection at the time of the Norwood procedure, pulmonary veins anomalously connected to a systemic venous site should be repaired before the Fontan procedure is completed (1).

During heart transplantation, the posterior walls of the recipient's atria with their venous connections (systemic and pulmonary) are normally left in situ and then sutured to the

donor's atria (2). In a recipient patient with anomalously connected pulmonary veins, an anastomosis must be made between the pulmonary venous confluence and the donor's left atrium.

The term *connection* refers to the anatomic attachment of the pulmonary veins (thus, normal connection means attachment of the pulmonary veins to the morphologic left atrium). The term *drainage* refers to the physiologic end point of pulmonary venous flow (normal drainage occurs when pulmonary venous flow travels into the systemic arterial circulation).

The current study was undertaken to determine 1) the prevalence of echocardiographically detected anomalous connection or drainage, or both, of the pulmonary veins in a large cohort of patients with hypoplastic left heart syndrome who underwent the Norwood procedure; and 2) the additive diagnostic role of Doppler color flow mapping to two-dimensional echocardiography and conventional Doppler study in precise definition of the connection and drainage sites of the pulmonary veins in hypoplastic left heart syndrome.

## Methods

Study patients. Between February 1987 and October 1990, 317 newborn infants with hypoplastic left heart syndrome underwent the Norwood procedure (that is, atrial

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septectomy and pulmonary artery to ascending aorta anastomosis with reconstruction of the aortic arch and placement of a 4-mm central or modified right Blalock-Taussig shunt). Two anatomic groups were included in this series: 1) 294 patients with aortic atresia (or severe aortic stenosis) with hypoplastic left ventricle; and 2) 23 patients with unbalanced complete common atrioventricular canal with a hypoplastic left ventricle. Patients with double-outlet right ventricle were specifically excluded from this study because of the known high prevalence of anomalous pulmonary venous connections in cases with heterotaxy syndrome and double-outlet right ventricle. During the study interval, no patient underwent heart transplantation and none was refused surgical palliation because of pulmonary venous anomalies.

**Echocardiographic examination.** Each patient underwent, as the primary preoperative diagnostic study, a comprehensive two-dimensional and Doppler echocardiographic examination performed by a technologist and supervised by an attending physician. Hewlett-Packard 77020 phased array ultrasound systems (Sonos 500 and 1000) with 5-MHz short- and medium-focus transducers were used. All studies were recorded on 0.5-in. (1.27-cm) VHS format videotapes and were available for retrospective off-line analysis. The echocardiographic examination included sequential scanning from subcostal, apical, parasternal and suprasternal windows. Blood flow patterns were assessed with pulsed continuous wave ultrasound, as well as Doppler color flow mapping in each patient. As a routine, two-dimensional echocardiographic imaging was performed first from all windows, followed by conventional Doppler study and color flow mapping. Each patient, therefore, was used as his or her own control to determine the additive role of color flow mapping with respect to two-dimensional echocardiography and conventional Doppler study in obtaining additional information on the precise sites of connection or drainage, or both, of pulmonary veins. All studies were initially interpreted by one of four observers and were reviewed retrospectively for the purpose of this study by one observer (M.A.S.). Operative notes were retrieved from the cardiothoracic surgery data base to determine the intraoperative observations.

Before hospital discharge, each patient underwent a comprehensive echocardiographic and Doppler study. In those with anomalous pulmonary venous connection, the pulmonary venous confluence-left atrial anastomosis was assessed and an attempt was made to detect the persistent vertical vein because the latter was usually not ligated. In patients with normal pulmonary venous connection but anomalous drainage, the surgically created atrial septal defect was assessed and an attempt was made to visualize the accessory vertical vein.

## Results

### Echocardiographic Findings

Two hundred ninety-seven patients had normal pulmonary venous connection and drainage. Anomalies of connec-

**Table 1. Clinical Characteristics of 20 Patients With Hypoplastic Left Heart Syndrome\* and Anomalous Pulmonary Venous Connection/Drainage Patterns**

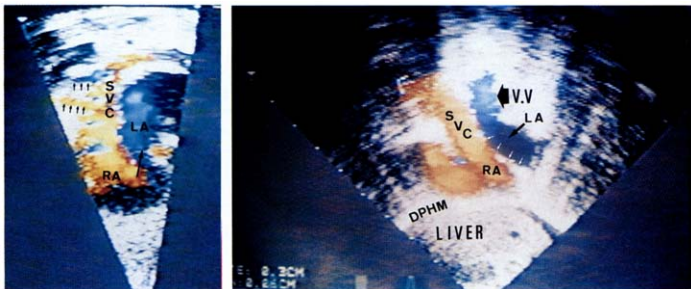
Case No.	Anomalous PV Pattern
1. Partial Anomalous Connection and Drainage	
1	Right PVs to the right SVC; normal left PVs connection to the LA
2	Right PVs to left SVC (left vertical vein) to the innominate vein and coronary sinus; normal left PV connection to the LA
2. Total Anomalous Connection and Drainage	
3 to 8	TAPVC to a confluence behind the LA; persistent left vertical vein to the innominate vein
9	Same as in Cases 3 to 8
10	Same as in Cases 3 to 8, except that the vertical vein drained to the coronary sinus
3. Normal Connection and Total Anomalous Drainage	
11 to 15	Normal PV connection to the LA; persistent vertical vein draining into the innominate vein
16 and 17	Same as in Cases 11 to 15, except that the vertical vein drained into the right SVC
18	Same as 11 to 15, except that the vertical vein drained into the coronary sinus
4. Non-1 Connection With Partial Anomalous Drainage	
19 and 20	Right PVs drained to the right of the deviated IAS

\*In all except Patient 9, who had unbalanced complete common atrioventricular canal, the hypoplastic left ventricle was associated with aortic atresia or severe aortic stenosis. IAS = interatrial septum; LA = left atrium; PV = pulmonary vein; SVC = superior vena cava; TAPVC = total anomalous pulmonary venous connection.

tion or drainage, or both, were detected by preoperative two-dimensional echocardiography and Doppler examination in 20 patients (6.3%). The views that best demonstrated the detailed anatomy of the pulmonary veins and their sites of connection or drainage, or both, were subcostal views (especially long-axis oblique and frontal views) and supracardiac frontal views. A morphologic left atrium was defined by whether the septum primum attached to the same side (that is, left) of the septum secundum, as well as by the presence of a morphologic left atrial appendage. Color flow mapping clearly demonstrated the direction of the flow in the pulmonary veins in all patients and confirmed the site of anomalous connection and drainage in each of the 20 cases. These patterns were confirmed intraoperatively and could be grouped as follows (Table 1).

1. **Partial anomalous connection and drainage (Cases 1 and 2).** In two patients (10%), the right pulmonary veins were connected and drained into the superior vena cava (Fig. 1). The left pulmonary veins were normally connected and drained into the left atrium.

2. **Total anomalous connection and drainage (Cases 3 to 10).** In eight patients (40%), all four pulmonary veins were connected not to the anatomic left atrium, but to a common venous confluence posterior to the left atrial cavity, with



**Figure 1 (left).** Case 1. Subcostal long-axis oblique view in a patient with hypoplastic left heart syndrome and partial anomalous pulmonary venous connection and drainage. The right pulmonary veins (small arrows) connect to the right superior vena cava (SVC), which drains into the right atrium (RA). The direction of the interatrial shunt at that point of the cardiac cycle (large arrow, blue color) is from the right to the left atrium (LA).

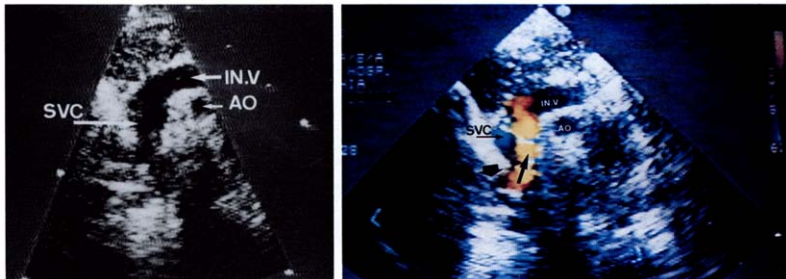
**Figure 2 (right).** Case 13. Subcostal sagittal view in a patient with normal connection and total anomalous pulmonary venous drainage. The interatrial septum (small arrows) was muscular, thick and intact. The four pulmonary veins connect normally to a chamber with a morphologic left atrial appendage. A vertical vein (V.V) drained the pulmonary vein into the innominate vein (flow away from the heart in blue color [arrowhead]). DPHM = diaphragm; LA = left atrium; RA = right atrium; SVC = superior vena cava.

presence of a vertical vein that drained into the innominate vein (seven patients) or the coronary sinus (one patient).

**3. Normal connection and total anomalous drainage (Cases 11 to 18).** In eight patients (40%), all four pulmonary veins were normally connected to the left atrium. However, in the

**4. Normal connection with partial anomalous drainage (Cases 19 and 20).** In two patients with leftward and posterior deviation of the superior attachment of the septum primum, the right pulmonary veins were connected to the

**Figure 3.** Case 17. Figure 4 demonstrating the value of Doppler color flow mapping in precise definition of the connection and drainage sites of the anomalous pulmonary veins. In this example, all four pulmonary veins connect to a vertical vein, which drains into the right superior vena cava (SVC) at the superior vena cava-innominate vein junction. **Left.** Suprasternal frontal (short-axis) view showing the superior vena cava-innominate vein (IN-V) junction. The anomalous connection site (of the vertical vein) is not clearly seen. **Right.** The same view with Doppler color flow mapping added showing the connection site (arrowhead) to the superior vena cava with the flow (red; arrow) toward the innominate vein. This pattern was seen in two patients. AO = hypoplastic aorta.



left atrium (that is, to the left of the septum secundum) but to the right of the superior attachment of the deviated septum primum and drained into the right atrium (Fig. 4).

**Pulmonary venous obstruction.** This was detected postoperatively in one patient with total anomalous connection and drainage. The left pulmonary veins were severely stenotic and required reoperation after the stage I Norwood procedure. The stenotic pulmonary veins were not detected by preoperative two-dimensional echocardiographic or conventional Doppler examination (including Doppler color flow mapping), probably because of lower blood flow velocity from the pulmonary veins to the pulmonary venous confluence before creation of a large atrial septal defect during the stage I Norwood procedure.

**Infradiaphragmatic connection or drainage.** Normal pulmonary venous connection with anomalous drainage to an infradiaphragmatic site was not found. Similarly, we did not find anomalous pulmonary venous connection to an infradiaphragmatic site among our series, which excluded cases of double-outlet right ventricle.

**Doppler color flow mapping.** The additive role of Doppler color flow mapping with respect to two-dimensional echocardiography and conventional Doppler ultrasound was clearly demonstrated in two settings: 1) detection of small accessory vertical veins, their course and site of narrowing (Fig. 3 and 5); and 2) detection of the drainage of the right pulmonary veins to the right of the septum primum in cases with leftward and posterior deviation of superior attachment of the septum primum (Fig. 4).

### Surgical Repair

In the two patients with partial anomalous connection and drainage into the right or left superior vena cava (pattern 1), the right pulmonary veins were not reconnected to the left atrium during the initial palliative procedure. In patients with total anomalous connection of the pulmonary veins (pattern 2), the pulmonary venous confluence was anastomosed to the anatomic left atrium during the Norwood procedure. In patients with normal connection and total anomalous drainage (pattern 3), the vertical vein was not ligated and the flow through it (examined by Doppler color flow mapping in a postoperative study) either stopped or became very small after surgical creation of a large atrial septal defect. In the two patients with a deviated septum primum (pattern 4), no intervention was required because the right pulmonary veins drained into what becomes the pulmonary venous chamber after completion of the modified Fontan procedure.

### Discussion

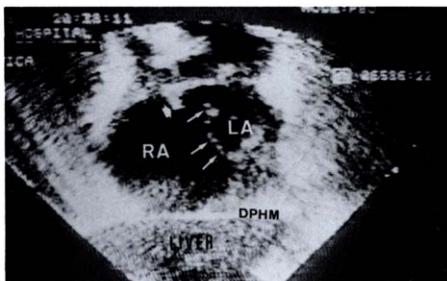
Hypoplastic left heart syndrome is the most common congenital cardiac malformation in which there is only one effective ventricle (1). Currently, there are two surgical procedures for its repair: 1) staged reconstructive repair, and 2) heart transplantation (2). Preoperative definition of the

detailed anatomy of the atria and their venous connections is important for the surgeon during either procedure to avoid the need for prolonged intraoperative surgical inspection with subsequent prolongation of the procedure.

**Anomalies of pulmonary venous connection/drainage in hypoplastic left heart syndrome.** In this series, echocardiographically demonstrable anomalies of pulmonary venous connection or drainage were found in 6.3% of our patients with hypoplastic left heart syndrome with normally aligned great arteries who underwent the stage I Norwood procedure. Most previous reports addressing this specific question were pathologic studies. Shone and Edwards (3) examined the association between mitral atresia (in hypoplastic left heart syndrome) and pulmonary venous anomalies in 29 cases. Five (17%) of these cases had anomalous venous connection or drainage. In one case, the left upper pulmonary vein connected anomalously to a vertical vein that drained into the left innominate vein (analogous to pattern 1 in our series). In a second case, all four pulmonary veins connected anomalously to two separate vertical veins that drained into the left innominate vein (analogous to pattern 2 in this series). In this latter case, the atrial septum was "absent." In two other cases, all four pulmonary veins connected normally to the morphologic left atrium, the foramen ovale was closed and a persistent vertical vein connecting the left upper pulmonary vein to the left innominate vein was detected in one case and assumed to be the probable collateral channel in the other case (analogous to pattern 3 in our series). In their last case, there was an associated cor triatriatum in which all four pulmonary veins connected to the accessory left atrial chamber and the true left atrium communicated through a patent foramen ovale with the right atrium. The investigators explained the development of anomalous connection of the pulmonary veins in three cases on the basis of obstruction to the blood flow from the left atrium (mitral atresia with premature closure of the foramen ovale). However, such an obstructive factor could not be found in the other two cases (one with a common atrium) to explain why the pulmonary veins connected anomalously.

**Role of abnormal foramen ovale formation in hypoplastic left heart syndrome.** Bharati and Lev (4) postulated that abnormal formation of the foramen ovale may be an underlying developmental factor in hypoplastic left heart syndrome. Among 230 autopsy hearts with this syndrome, they found 34 hearts (15%) with a small patent foramen ovale (3 mm) and 2 hearts (0.9%) with a congenitally closed foramen. However, 84% of the autopsy hearts had adequate interatrial communication, a secundum atrial septal defect (105 hearts), a wide foramen ovale (88 hearts) and a common atrium (1 heart). Their own data, therefore, argue against such a cause and effect relation as the primary developmental phenomenon at least in the majority of cases of hypoplastic left heart syndrome. They mentioned only one case (0.4%) with anomalous connection (the left upper pulmonary

**Figure 4.** Case 19. A, Subcostal long-axis oblique view in a patient with leftward and posterior deviation of the superior attachment of the septum primum (arrows), which is far away from the septum secundum (arrow-head), B, With Doppler color flow mapping added, the flow from the right pulmonary veins (large arrow) is directly draining into the right atrium (RA) because these veins connect to the left of the septum secundum (arrow-head), but to the right of the deviated septum primum (small arrows). DPHM = diaphragm; LA = left atrium.



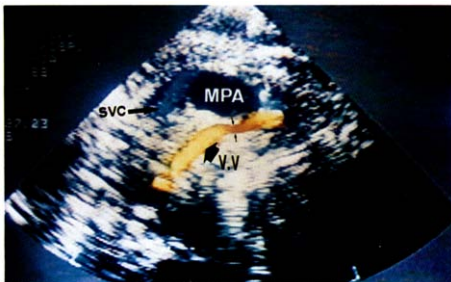
vein connected to the left superior vena cava, which connected to the coronary sinus).

The apparent difference between the prevalence of pulmonary venous anomalies noted in our series and that reported by Bharati and Lev (4) may in part be related to the difference in the study design. They did not specifically

comment on the pulmonary venous connection patterns in their cases.

The presence of a small or congenitally closed foramen ovale is conceivably the underlying factor for development of some of these anomalous pulmonary venous patterns. If aortic or mitral atresia (or severe stenosis), or both, were

**Figure 5.** Case 15. Suprasternal frontal view showing the winding course of the vertical vein (V.V.) in the superior mediastinum with color flow mapping. Note the area of mild narrowing (small arrows) as the vertical vein passes between the left pulmonary artery and left main stem bronchus. MPA = main pulmonary artery; SVC = superior vena cava.



truly the primary developmental event in hypoplastic left heart syndrome (1), the presence of a restrictive foramen ovale very early during intrauterine life might have led to failure of incorporation of the common pulmonary vein into the anatomic left atrium, with persistence of one of the cardinal veins as a collateral channel draining the pulmonary venous flow into a systemic site as in our Patients 3 to 10 (Table 1). If the foramen ovale became restrictive during a later phase of development after connection of the pulmonary veins to the left atrium, the only developmental consequence might have been persistence of one of the cardinal veins (accessory vertical veins) as a collateral channel to bypass the obstruction of pulmonary venous flow through the left atrium as in our Patients 11 to 18 (Table 1) (3). However, these theories may oversimplify the developmental cause of anomalous pulmonary venous patterns in hypoplastic left heart syndrome (for example, why should a patient with hypoplastic left heart syndrome and a common atrium (3) develop total anomalous pulmonary venous connection?).

The anatomy of the atrial septum and its variations in hypoplastic left heart syndrome was previously reported in detail in two reports from this Institution (5,6). Several other pathologic reports (7-9) included sporadic cases of anomalous pulmonary venous connections in hypoplastic left heart syndrome.

**Surgical implications.** Although lack of one of these anomalous patterns (in the absence of pulmonary vein obstruction) does not affect the outcome of the patient after the stage I Norwood procedure, it creates a hemodynamic problem with completion of the Fontan procedure later on. In patients with anomalous connection (patterns 1 and 2), the pulmonary veins would be connecting to and draining into the systemic venous channel (that is, into the lungs). In patients with anomalous drainage (pattern 3), a persistent vertical vein (if not ligated) may remain as a potential shunting site between the systemic and pulmonary venous channels after completion of the Fontan procedure. In one patient, a right to left shunt through such a vertical vein was detected during postoperative cardiac catheterization and coil occlusion was performed because of systemic oxygen desaturation. Finally, a false diagnosis of anomalous connection in a patient with a normal connection and anomalous drainage (pattern 3) may result in prolongation of the operative procedure as the surgeon tries to define precisely the pattern of pulmonary venous connection to the left atrium posterior to the heart when all that is needed is the creation of an atrial septal defect.

**Role of Doppler color flow mapping in echocardiographic diagnosis.** Several reports (10-15) have described the use of two-dimensional echocardiography and conventional Doppler examination (with or without contrast injection) in the diagnosis of partial or total anomalous pulmonary venous connections. Both techniques, however, have met with difficulties and errors in precise diagnosis of the anatomic sites of connection and drainage, especially in the setting of complex congenital heart lesions (14). Some advantages of

adding color flow mapping to these two techniques have been described (16). Our study demonstrated the value of color flow mapping in this setting, which showed more variable patterns of anomalous connection and drainage of the pulmonary veins than those found in isolated forms with an otherwise structurally normal heart.

The additive role of Doppler color flow mapping to two-dimensional echocardiography and conventional Doppler study was clearly demonstrated in this study in two situations. The first is the detection and definition of the exact course of small accessory vertical veins (Fig. 5) which, during their course in the superior mediastinum, may pass between a pulmonary artery branch and main stem bronchus where they may be difficult to follow by two-dimensional echocardiography or conventional Doppler study alone. The addition of color flow mapping in these cases enhanced the precise definition of the course of the vertical vein and its connection site (Fig. 3). The second is detection of the drainage of the right pulmonary veins to the right of the atrial septum in cases with leftward and posterior deviation of the superior attachment of the septum primum. In these cases, although the right pulmonary veins connect normally to the posterior wall of the left atrium, color flow mapping clearly demonstrated the drainage of their flow through the gap between the septum secundum and the superior attachment of the septum primum (Fig. 4), which would be difficult to detect by other Doppler techniques.

**Limitations of the study.** Although there are no exclusion criteria at our institution for offering the Norwood procedure (except in cases of severe pulmonary valve stenosis or lethal chromosomal anomaly), it is possible that a predominance of complicated cases are referred to our center. Thus, our estimate of the prevalence of anomalous pulmonary venous connection or drainage, or both, may slightly overestimate the true prevalence.

Second, because we used intraoperative observation as our reference standard, our prevalence data are only approximate (autopsy, clearly the most accurate way of distinguishing the connection pattern of the pulmonary veins, was not available for most of the study group).

Finally, it may not be possible to avoid diagnostic error in the few cases in which the morphologic left atrial appendage (which defines the true morphologic left atrium) is hard to define echocardiographically. In such cases, it is difficult to be certain whether the chamber to which the pulmonary veins are connected is a left atrium or a pulmonary venous confluence.

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