Surgical Treatment of Partial Anomalous Pulmonary Venous Drainage

A Long-Term Follow-up Study

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SUMMARY

Fourteen children who had been operated on for partial anomalous pulmonary venous return from the right lung to the vena cava or right atrium have been submitted to a follow-up study 1 to 9 years after surgery. This included clinical, hemodynamic, and angiographic data. All patients were doing well and had normal exercise tolerance at the time of follow-up examination. Electrocardiograms and chest roentgenograms had returned to normal in 10 and were improved in three. Hemodynamic and angiographic data demonstrated perfect correction in eight. Of the remaining six, three had an obstructed superior vena cava (SVC) with collateral pathways to a persistent left superior vena cava in two and to the inferior vena cava in one. Three had significant pressure gradients between the SVC and the right atrium, two with a small right-to-left shunt from the SVC to the left atrium, and one with an additional small left-to-right shunt. None had obstruction to the pulmonary venous return. These complications occurred exclusively in cases in which at least one anomalous vein drained high into the superior vena cava. Enlargement of the SVC with a patch of pericardium gave good results in three patients with this type of anomalous drainage but did not prevent obstruction or severe narrowing of the SVC in two other cases. It is concluded that the present corrective technics are not adequate for cases in which an anomalous vein drains high in the SVC, especially in the presence of a left superior vena cava.

Additional Indexing Words:
Congenital heart disease Sinus venosus defect Atrial septal defect
Superior vena cava obstruction

PULMONARY venous drainage into the right heart is a frequent anomaly and has been found in 0.7% of routine autopsies. The most common variety involves draining of veins from the right lung into the superior vena cava (SVC) or right atrium (RA) or both. It may remain unsuspected throughout life. However, when it is associated with an atrial septal defect (ASD), symptoms will often occur during childhood, that is, earlier than in uncomplicated ASD, especially when the left-to-right shunt exceeds 50%. This group of children require surgical correction. A number of approaches have been devised, but basically four types of operations are used:

1. A Teflon or Ivalon patch is sutured around the entry of the anomalous veins and the ASD in such a way as to redirect the flow from the pulmonary veins into the left atrium (LA) through the ASD.

2. When the anomalous veins drain directly into the RA, the atrial septum is mobilized and sutured to the lateral wall of the right atrium (RA) in front of the pulmonary veins.

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Received June 22, 1971; revision accepted for publication September 10, 1971.
3. The anomalous vein is cut off and reimplanted into the LA or another pulmonary vein, especially if it drains high into the superior vena cava.6-11

4. The SVC is partitioned into two separate channels, one draining the anomalous veins into the LA through the ASD, the other draining the systemic venous return into the RA.6, 9, 12

All types of operations have generally given clinically satisfactory results; however postoperative hemodynamic data are scanty in the literature and always for a short follow-up (a few weeks to a few months).1, 9, 10, 12 Angiographic data are lacking. This has prompted us to undertake a follow-up study of our cases, including hemodynamic and angiographic evaluation.

Methods

Group Studied

From 1961 to 1970, 17 patients with partial anomalous pulmonary venous drainage were operated on in this hospital. Fourteen patients (cases 1 to 14) could be brought back to the hospital for reevaluation. Thus the delay from operation to reevaluation ranges from 1 to 9 years. Patients' ages at operation ranged from 5 to 15 years. Figure 1 shows the anatomic aspect of the anomalous pulmonary veins and the atrial septum as observed in our patients. In summary, nine patients (upper row) had at least one anomalous

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Figure 1

Schematic drawings according to Snellen,1 showing the types of anomalous pulmonary venous return in this study. The drawings show right and left atrium, atrial septum with the two types of defect, superior and inferior vena cava, coronary sinus, and the pulmonary veins. (Upper row) At least one anomalous vein drains high into the superior vena cava. (Lower row) Drainage is low into the superior vena cava, directly into the right atrium, or into the inferior vena cava close to the right atrium.

Abbreviations: APV = anomalous pulmonary veins; SVC = superior vena cava; IVC = inferior vena cava; RA = right atrium; ASD = atrial septal defect, secundum type; SV = sinus venosus type of defect; LSVC = persistent left superior vena cava; AS = atrial septum.
Figure 2

(A) Diagram of external appearance of heart with partial anomalous pulmonary venous return to superior vena cava (S.V.C.). (B) Appearance of partial anomalous venous return to superior vena cava with atrial septal defect as seen through the right atrium (RA). (C) The superior vena cava has been partitioned and the pulmonary venous return directed to the left atrium through the atrial septal defect. (D) Superior vena cava enlarged with a pericardial patch.

Abbreviations: P.V. = pulmonary veins; Ao. = aorta; P.A. = pulmonary artery.

vein draining high into the SVC (just below the azygos vein); three of them had an intact atrial septum; three had a high (sinus venosus) ASD; and three had a high defect and a persistent left superior vena cava (case 9 was drawn separately because the entire right lung drained into the
right heart). Five patients (lower row) had anomalous drainage into the SVC, close to the RA, directly into the RA or, in case 6, into the inferior vena cava (IVC).

**Operative Technic**

Partitioning of the SVC with a longitudinal suture starting above the highest pulmonary vein and redirecting the pulmonary venous flow through the ASD into the LA was used in 10 patients (cases 1, 2, 4, 5, 8, 10, 11 to 14); when the atrial septum was intact, a surgical defect was created. In the two cases (cases 3, 7) in which the anomalous drainage was into the RA, the medial edge of the atrial septum was sewn to the wall of the RA, anteriorly to the pulmonary veins (atrioseptopexy). In case 6, a partitioning of the IVC was done. In one case (case 9), in which partitioning of the SVC proved difficult, a pericardial patch was sewn around the orifices of the anomalous veins and the ASD, separating the systemic and pulmonary venous returns. As the partitioning procedure reduces the diameter of the SVC, it appeared advisable to enlarge this vessel with a pericardial patch (fig. 2). This has been done since 1967 in five cases (cases 10 to 14).

**Follow-up Study**

This included clinical examination, ECG, chest X-rays, phonocardiogram, and right heart catheterization. The catheter study was devised to collect the following data: pressure measurements in the right heart, especially measurement of the gradient between RA and SVC, and pulmonary capillary wedge pressure in the lobe from which the vein was redirected. This was obtained in only six cases, as it sometimes proved difficult to

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**Table 1**

**Clinical, ECG, and X-Ray Data**

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<th>PO</th>
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<th>X-rays</th>
<th>Preop</th>
<th>PO</th>
<th>Vascular markings</th>
<th>CTR</th>
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Abbreviations: + = mildly symptomatic; ++ = moderately symptomatic; URI = repeated upper respiratory tract infection; WNL = within normal limits; IRBB = incomplete right bundle-branch block; RVH = right ventricular hypertrophy; RAD = right axis deviation; ↑ = mildly increased; ↑↑ = markedly increased; CTR = cardiothoracic ratio; N = normal.
Table 2

### Hemodynamic Data

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<th>L-R shunt (%)</th>
<th>Operation Type</th>
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<th>Pulmonary artery pressure (mm Hg)</th>
<th>Mean wedge PO</th>
<th>SVC PO</th>
<th>Gradient SVC-RA (mm Hg)</th>
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Abbreviations: SVC = superior vena cava; RA = right atrium; IVC = inferior vena cava; ASD II = atrial septal defect, secundum type; PO = postoperative.
enter an upper lobar artery. To detect any residual left-to-right shunt, serial O₂ saturations from SVC to pulmonary artery (PA) were obtained by using a Wood's cuvette oximeter, and dye-dilution or hydrogen curves or all three. Finally, the pulmonary venous return and the SVC-to-RA junction were visualized by injection of diatrizoate (Hypaque) into the right PA and SVC, respectively. These angiograms were recorded on an Elema-Schonander biplane serigraph.

Results

Clinical, ECG, and X-Ray Data

These are summarized in table 1. All patients were mildly (+) or moderately (++) symptomatic, that is had dyspnea and fatigue on exercise, prior to the operation. Eight had repeated, sometimes severe, upper respiratory tract infections (URI). At the time of the follow-up study, all were asymptomatic and participating in sports with normal exercise tolerance. Preoperative ECGs showed incomplete right bundle-branch block (IRBB) in seven (five with ASD of some type, and two with intact septum) which persisted in all after operation. Nine showed evidence of right ventricular hypertrophy (questionable in one), which disappeared completely in seven and decreased in the other two.

Minor atrial arrhythmias not seen before operation were found in five cases at the time of follow-up study: Three of these patients had coronary sinus rhythm, one had a wandering pacemaker, and one had a nodal rhythm competing with the sinus rhythm.

Figure 3

Case 10. Angiogram after injection into the left innominate vein (lateral view), showing a good-sized superior vena cava after surgery (no significant pressure gradient between S.V.C. and R.A.). Abbreviations: Az. = azygos vein; R.V. = ventricle. Other abbreviations same as in figures 1 and 2.
(interference dissociation). Preoperative chest roentgenograms showed increased vascular markings in all with a cardiothoracic ratio (CT) ranging from 0.49 to 0.58. The chest X-rays, obtained at the follow-up study, were normal in 13 patients with CT ratios ranging from 0.41 to 0.50. The only patient who had an associated VSD closed at operation had a CT ratio of 0.54, 2 years after surgery. In six cases, systolic clicks were heard and documented by phonocardiograms; these had not been mentioned preoperatively.

**Hemodynamic Findings**

These are summarized in table 2. Before operation, left-to-right shunts ranged from 40 to 75%. Six patients had a systolic pulmonary arterial (PA) pressure slightly elevated before operation (27 to 36 mm Hg). After operation, all had normal PA pressures except for a slightly elevated value in case 6 (31/15 mm Hg).

**Pulmonary Venous Return**

Mean pulmonary capillary wedge pressure from the lobe where the vein was redirected was obtained in six cases. It was within normal limits in five (5 to 12 mm Hg) and slightly elevated in one (case 6; 17 mm Hg); in this patient the entire right lung had drained into the IVC prior to corrective surgery.

Figure 4

*Case 9. The catheter is placed into the right innominate vein (R.I.). The superior vena cava (S.V.C.) is obstructed above the azygos vein. The systemic venous return is directed toward a persistent left superior vena cava by collateral pathways, mainly through the jugular vein (J.V.) and inferior thyroid vein (I.T.V.). Abbreviations: Sc.V. = subclavian vein; L.S.V.C. = left superior vena cava; C.S. = coronary sinus; P.E.V. = phrenoepticardic vein.*
Patency of the Superior Vena Cava

The SCV was patent without narrowing in eight cases. In cases 8 and 9 it was occluded, and in case 11 it was obstructed. In cases 8 and 9 a large left superior vena cava drained into the coronary sinus and an abnormally small right superior vena cava was noted at operation. The mean SVC pressure was 8 and 12 mm Hg in these cases. Patient 11 had no left superior vena cava and a SVC pressure of 20 mm Hg. All three patients had at least one pulmonary vein draining high into the right SVC. Three additional patients (cases 2, 4, and 14) were found to have significant pressure gradients between the RA and the SVC of 10, 9, and 11 mm Hg, respectively. All three had an anomalous pulmonary vein draining high into the SVC. The mean SVC pressure in these three cases was about 15 mm Hg.

Shunts

Two patients (cases 2 and 4) had a slight right-to-left shunt at time of follow-up as demonstrated by oximetry and dye-dilution curves, estimated to be 7 and 15%, respectively. In addition, one patient (case 2) had a mild left-to-right shunt (18%, by oximetry). In this case, the catheter entered the LA and left ventricle (LV) from the SVC. An additional patient (case 1) was found to have a minimal

Figure 5

Case 11. Posteroanterior view. The catheter is placed into the left innominate vein (L.I.) The superior vena cava is obstructed above the azygos vein, which had been ligated during surgery. The main collateral pathway to the inferior vena cava is by the accessory hemi-azygos vein (A.H.Az.V.), that is reached through the highest intercostal vein (H.I.C.V.). Minor collateral pathways are through the internal mammary vein (I.M.V.) and the phrenoepticardic vein (P.E.V.). (See figure 6 for lateral view.)
left-to-right residual shunt detected only by hydrogen curve.

**Angiographic Data**

Figure 3 shows good surgical result in a case in which the anomalous drainage was high into the SVC. Partitioning resulted in a good-sized SVC and a pulmonary vein draining through the newly created channel into the left atrium. In this case the SVC had been enlarged by a pericardial patch.

In cases in which the SVC was obstructed, rich collateral pathways were demonstrated by the angiograms. In the two cases in which a left superior vena cava existed the collaterals between the jugular veins directed the blood toward this vessel (fig. 4). In case 11, where no left superior vena cava existed, a large collateral pathway entered the hemiazygos-azygos system through the innominate vein and the highest intercostal vein to end up in the inferior vena cava (fig. 5 and 6). The azygos vein had been ligated during operation. In case 4 a right-to-left shunt could be well demonstrated: From an injection into the SVC, opacification of the azygos vein, the anomalous pulmonary vein, and the two channels resulting from the partitioning occurred (fig. 7). Opacification of the LA and LV was seen 2 sec after the injection. (This patient has a 15% right-to-left shunt by oximetric data).

**Discussion**

Surgical correction of partial anomalous pulmonary venous connection with (or without) ASD is a more difficult procedure than closure of an ASD alone. The potential hazards are: (1) incomplete correction with persistent left-to-right shunt, (2) obstruction

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**Figure 6**

*Case 11. Lateral view. See figure 5 for description and posteroanterior view.*

*Circulation, Volume XLV, January 1972*
Figure 7

Case 4: The catheter is placed into the superior vena cava. Injection of contrast material opacifies the azygos; the S.V.C. below the azygos vein is divided into anterior and a posterior channel. The anterior, smaller one is in connection with the right atrium. (Pressure gradient of 9 mm Hg between R.A. and S.V.C.). The posterior one, in connection with the left atrium and the pulmonary veins, is opacified through a small leak in the suture line. C. = channel. Other abbreviations as in figures 1 to 6.

of the redirected pulmonary veins, and (3) complete or partial obstruction of the SVC.8, 9, 10, 12

Bailey’s atrioseptopexy has been widely used6, 10, 11 in cases in which the anomalous veins drain into the RA. This operation gives excellent results and avoids the insertion of foreign material into the atrium. No complication has occurred in our cases with this type of drainage.

When one anomalous vein drains high into the SVC, usually just below the azygos vein, considerable narrowing of the SVC may occur as a result of surgery, whether the Teflon patch technic8 or the longitudinal partitioning procedure12 is used. Schuster and associates,8 therefore, recommended the enlargement of the SVC by means of a patch of pericardium. They operated on five patients with this technic with good results in four, and transient signs of obstruction of the SVC in one. Postoperative catheterization was not done, and it is possible that the subsequent disappearance of the signs of SVC obstruction was due to the establishment of collaterals rather than to recanalization of the vein.

Various other approaches have been used to cope with the problem of the high anomalous pulmonary vein: section and reimplantation into the LA or another pulmonary vein is advocated by Risch and Hahn;10 ligation of the high draining vein has been done,6 and in

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one case a right upper lobectomy. Mustard and Brock and Ross recommended leaving untouched small anomalous veins that drain high into the SVC. This would leave the patient with a small, probably hemodynamically insignificant shunt.

In our series, all patients are doing well clinically. They have become asymptomatic and have normal exercise tolerance. Electrocardiograms and chest X-rays have returned to normal in almost all cases. The appearance of a systolic click after surgery was surprising to us, and we have found no firm explanation for it. No correlation exists between this sound and the type of anomaly or type of operation; an extracardiac origin cannot be excluded.

Hemodynamic and angiographic data are, on the other hand, partly in contrast with these good clinical results. Although near-normal angiograms were obtained in eight patients, 2 patients had evidence of complete obstruction of the SVC and four had narrowing of this vessel with significant pressure gradients. The site of entry of the anomalous veins is most important in this regard. No complications occurred when the anomalous veins drained into the RA or into the vena cava close to the RA, regardless of the type of surgery performed. On the other hand, when part of the anomalous drainage was high into the SVC, the partitioning procedure without enlargement of the vena cava led to a significant narrowing or occlusion of this vessel in four of the five cases. When a pericardial patch was used in order to enlarge the SVC (fig. 2), good results were obtained in three out of the five cases (fig. 3). In the two remaining cases, complete obstruction or severe narrowing could not be prevented by this procedure. Whether this is due to early thrombosis or constrictive scarring is uncertain. Experimental data from our laboratory and from other workers make a thrombotic process more likely. In dogs, replacement of a segment of the SVC by a piece of pericardium results in a high percentage of thrombotic occlusion. This can be prevented by anticoagulants.

When one considers that a single anomalous pulmonary vein (without ASD) may remain unsuspected throughout life; it seems advisable to leave such a vein untouched during surgery when it drains high into the vena cava. The risk of narrowing and occlusion resulting from the present types of plastic surgery on the SVC is thus reduced.

Ligation of these high veins does not seem advisable: On the one hand, edema of the pulmonary segments draining to these vessels is possible in the immediate postoperative course, and the other blood draining to these occluded veins will eventually be directed to the cava through the bronchiozygous system, thus restoring the left-to-right shunt.

The mild right-to-left shunt from the SVC to the LA observed in two cases is a consequence of the narrowing of the vena cava. As pressure builds up in the SVC, exceeding LA pressure, systemic venous blood may gain access to the anomalous pulmonary vein through a leak in the suture line and from there to the LA.

We believe the following conclusions should be drawn from the present study:

1. Surgical correction of partial anomalous pulmonary venous drainage gives good results when the drainage is into the RA or the vena cava close to the RA.
2. A definite risk of narrowing and occlusion of the SVC exists when the procedure is used to redirect an anomalous vein draining far from the RA. Even if the SVC is enlarged with a pericardial patch, especially in the presence of a L SVC, the RSVC then being narrower than usual. Thus, if this pulmonary vein is small, redirection of its flow should not be attempted with the present technics.
3. Clinical examination alone is not enough to evaluate the results of the operation. Hemodynamic and angiographic data must be obtained to ascertain the validity of a surgical procedure aimed at the correction of partial anomalous pulmonary drainage. Children with almost complete obstruction of the SVC seem to tolerate this anomaly rather well.

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Surgical Treatment of Partial Anomalous Pulmonary Venous Drainage: A Long-Term Follow-up Study
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Circulation. 1972;45:159-170
doi: 10.1161/01.CIR.45.1.159

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