Univentricular Heart of Right Ventricular Type with Double or Common Inlet

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SUMMARY Seventeen cases are described in which both atria connect directly to a chamber with right ventricular characteristics. The atria connected through separate atrioventricular valves in six hearts and a common valve in 11. All hearts had a posterior rudimentary chamber. The septum which separated it from the main chamber was directed to the crux of the heart. Ten hearts were from patients with atrial situs solitus and seven from patients with atrial situs ambiguous. Arterial connections were concordant in three cases, had a double outlet from the main ventricular chamber in nine and single outlet of the heart in five. The patent artery always arose from the main chamber, with pulmonary atresia in three and aortic atresia in two.

This and other studies indicate that double inlet atrioventricular connection does not predict the morphology of the main chamber. Although usually associated with a main chamber of left ventricular type, it may also be associated with a main chamber having right ventricular characteristics. Both types should be considered as univentricular hearts; the posterior chamber in hearts of right ventricular type are analogous to the anterior chamber in univentricular hearts of left ventricular type and are a rudimentary chamber rather than a hypoplastic ventricle. In the right ventricular form of univentricular heart, the trabecular zone of the rudimentary chamber is of left ventricular type.

HEARTS HAVE BEEN DESCRIBED IN WHICH BOTH ATRIA connect through either separate atrioventricular valves or a common valve with a chamber having the trabecular characteristics of a right ventricle. In some of these hearts the valve receiving chamber is the sole ventricular chamber, and is termed the "single right ventricle." However, occasionally an additional chamber with left ventricular trabecular pattern is present. This anomaly has been given a variety of names.

It would appear that these hearts satisfy the criteria used by Van Praagh et al. and Lev and his colleagues for inclusion in the category of "single or common" ventricle, regardless of the presence of an additional chamber, since both atria communicate directly with the same ventricular chamber. Yet the nature of the second chamber, when present, is uncertain, and no such hearts were included in the initial description of single right ventricle. This uncertainty relates to the lack of a precise definition of a ventricle. In this report, we describe 17 hearts, studied at autopsy, in which both atria connected through two valves or a common valve, principally with a chamber having right ventricular trabecular pattern. We describe how to diagnose this defect in life and suggest criteria for its classification.

Materials and Methods

During examination of autopsy specimens from five centers, we discovered 17 hearts in which both atria communicated via two separate valves or a common atrioventricular valve, with a ventricular chamber with right ventricular characteristics. Six were from the Hospital for Sick Children, London; five were from the Brompton Hospital, London; three were from the Royal Liverpool Children's Hospital; two were from Newcastle General Hospital; and one was from Killingbeck Hospital, Leeds. Each heart was examined morphologically, and four hearts were sectioned to identify the position of the conducting tissue (Wilkinson JL, Dickinson DF, Smith A, Anderson RH: submitted for publication). During our study we found that both valves did not have to be committed entirely to the chamber of right ventricular morphology. We also noted a spectrum, as described by others, between these hearts and hearts with straddling mitral valves. In this study, therefore, we included only hearts in which one complete valve, and more than half a straddling valve, were committed to the chamber with right ventricular trabecular pattern. In some of the hearts with a common atrioventricular orifice, the entire orifice was committed to the main chamber. These were included in the series. In other hearts we could discern distinct mitral and tricuspid components of the common valve by virtue of the attachments of the valve leaflets to the septum. In these latter specimens, the mitral component was hypoplastic, but was committed to a hypoplastic left ventricular chamber. These hearts were not included in the univentricular category. The term univentricular is presently used, rather than "single" or "common" ventricle, since there is no consensus on the correct way to use the latter terms. The degree

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of straddling was assessed by viewing the relationships of valve and septum from the atrial side. When possible, the angiograms and catheter data of the patients were examined retrospectively. One of the hearts examined at autopsy was from a patient in whom the diagnosis had been made at catheterization. In all the other cases the defects were unsuspected during life.

Results

The features of each heart are given in table 1.

Atrial Situs

Ten cases showed atrial situs solitus and seven had situs ambiguous. Of the latter, two had left isomerism and five had right isomerism.

Atrioventricular Connection

In all cases the atrioventricular connection was (by selection of the cases) double inlet to a chamber with right ventricular trabecular pattern. Double inlet describes the type of connection of the atria to the ventricular mass. There was, however, variation in the mode of connection. In six cases the atria were connected via two atrioventricular valves, and in 11 the atria communicated with the ventricle via a common atrioventricular valve. In 15 hearts, the two atrioventricular valves or common valve communicated entirely with the right ventricular chamber. In one heart the right atrioventricular valve orifice straddled a septum; in another the left valve orifice straddled.

Morphology of Atrioventricular Valves

In the six cases with two atrioventricular valves, both valves possessed prominent leaflets facing each other in the valve receiving chamber. However, there were variations in papillary muscle pattern in that one of the valves took prominent muscles from the trabecula septomarginalis, as is typical for the normal tricuspid valve. This valve was right-sided in four hearts with left-sided rudimentary chambers, and left-sided in two hearts with right-sided rudimentary chambers. In one of the four hearts with left sided rudimentary chamber, the left valve straddled the septum, and was 85% committed to the main chamber (fig. 1). The papillary muscle arising from the rudimentary chamber was single and had a "mitral" pattern (fig. 1B). The straddling portion of the valve was also attached to a single papillary muscle, but this in turn arose from the trabecula septomarginalis. In the other hearts with left-sided rudimentary chambers, the left valve arose entirely from the main chamber and had a mitral pattern. In the hearts with right-sided
Table 1.

<table>
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<tr>
<th>Case</th>
<th>Situs</th>
<th>AV Valves</th>
<th>Rudimentary chamber</th>
<th>Position of chamber</th>
<th>Arterial connections</th>
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<td>2 valves</td>
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<td>Left</td>
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<td>Aorta anterior and left</td>
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<td>and 85% to main chamber</td>
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rudimentary chamber, the right valve straddled the septum in one by 60%, resembling a mitral valve with left-sided rudimentary chamber. In the other case, the right valve arose entirely from the main chamber and had paired papillary muscle groups, with one group attached to the trabecula septomarginalis (fig. 2). In the cases with common atrioventricular valves, all valves had the leaflet pattern of a common valve; at least four leaflets were recognizable (figs. 3 and 4). The papillary muscles were similar to common valve papillary attachments. In one case, a small, mitral-type single papillary muscle arose from a left-sided rudimentary chamber, but the common valve orifice was entirely committed to the main chamber. In the other 10 cases all papillary muscles arose from the main chamber. In two hearts with very small rudimentary chambers, the left-sided papillary muscles arising from the main chamber were hypoplastic.

Morphology of Main Ventricular Chamber

In each specimen, the valve-receiving chamber exhibited coarse trabeculations and a prominent trabecula septomarginalis on the posterior septal surface (fig. 4). The limbs of the trabecula septomarginalis in all but two instances cradled the inferior rim of a septal defect communicating with a posterior rudimentary chamber (fig. 4).
Rudimentary Chamber

In each case, we identified a rudimentary chamber lying posterior to the main chamber of right ventricular type. The rudimentary chamber varied in size; in two cases it was only a slit-like cavity. In the other 15, it had fine trabeculations and a smooth septal surface; that is, the morphological characteristics of the trabecular zone of the left ventricle. This rudimentary chamber lay to the left of the main chamber in 14 cases (fig. 1) and to the right in three cases (fig. 2). In three cases the aorta arose from a left-sided rudimentary chamber, i.e., it had a direct arterial connection...
(fig. 3). In the remaining hearts no artery arose from the rudimentary chamber and no inlet portion was present, and the chamber had only a trabecular zone of left ventricular type (figs. 1, 2 and 4).

The septum separating the rudimentary chamber from the main chamber was posterior and always ran to the crux cordis. Its position was marked on the external surface of the heart by the posterior descending coronary artery (figs. 1, 2 and 4). In five cases the rudimentary chamber was not recognized on first examination, and was discovered only when an incision was made in the ventricular wall to the left of the posterior descending coronary artery, or when preparing the hearts for histological examination.

**Arterial Connections**

In three cases the aorta arose from the rudimentary chamber of left ventricular type and the pulmonary artery from the main chamber (fig. 3). This represents a concordant arterial connection, and the external relation of the arteries was for the aorta to be posterior and to the right of the pulmonary artery, as in the normal heart. One of the three cases showed severe isthmal hypoplasia, and the outlet to the aorta was barely probe patent. In nine cases, both the aorta and pulmonary artery arose from the main chamber; two of these had infundibular pulmonary atresia (fig. 1). In the remaining five cases only a single patent artery arose from the heart, always from the main chamber. In three cases, the patent artery was the aorta. There was pulmonary atresia, and the ventricular origin of the atretic pulmonary remnant could not be recognized with certainty. The remaining two cases showed aortic atresia, with the single outlet a pulmonary artery. Both these cases showed variation in ventricular morphology compared with the remaining cases (fig. 5). The rudimentary chamber was very small. The interventricular communication was identified in one heart posterior to the trabecula septomarginalis. In the other heart, the rudimentary chamber was identified only as a blood-filled cavity in the posterior wall of the ventricle (fig. 5B). In both hearts the aortic rudiment was posterior and to the right of the pulmonary trunk. In neither was it in potential communication with any chamber in the ventricular mass.

**Angiographic Correlations and Clinical Consideration**

In only one case was the diagnosis made during life from biplane cineangiography. In this case a common atrioventricular orifice opened into a triangular, rather heavily trabeculated chamber, and both great arteries arose from the same chamber. Posteriorly, a small pouch was outlined by contrast medium from the main chamber which was very slowly cleared. The chamber still could be seen several frames after the main chamber had emptied (fig. 6).

Angiocardograms were available from 11 of the other cases, but were inadequate for diagnosis.

**Discussion**

The cases we describe of two atrioventricular valves or a common atrioventricular valve opening entirely
Diagnostic Considerations

Realizing that cases could exist of a chamber with right ventricular characteristics receiving the atrial inputs, but also possessing a second posterior chamber, we have begun to recognize the anomaly in life. The small chamber was identified by angiocardiography in case 10. Had it not been demonstrated, it could well have gone unrecognized during autopsy. In the cases studied histologically, the posterior chamber had been unrecognized until blocks were removed for microscopic examination. These considerations suggest that the anomaly of double or common inlet to a chamber with right ventricular trabecular pattern may be more common than previously realized. Recognition of the rudimentary chamber is probably the most reliable means of distinguishing the anomaly during life, since differentiation of a chamber with right ventricular characteristics from a chamber with indeterminate characteristics, as in a univentricular heart without a rudimentary chamber, may be particularly difficult. In such cases the investigator should try to obtain angiocardiographic views which profile the posterior septum and rudimentary chamber, if present. If the rudimentary chamber supports a great artery (the aorta in three of our cases), it will be recognized more easily by retrograde selective injection of the chamber. These techniques should make possible the identification during life of univentricular heart of right ventricular type with rudimentary chamber, except when the chamber is minute and virtually obliterated. The demarcation of this type of univentricular heart with double or common inlet is important for subsequent surgery.
Surgical Considerations

Many of the cases we describe probably could not be corrected surgically, because of the frequent association of situs ambiguous and its concomitant venous anomalies, a common atrioventricular valve and either aortic or pulmonary atresia. In all the hearts we studied, the rudimentary chamber of left ventricular type was insignificant in terms of function, and can probably be ignored for the purposes of correction. Hemodynamic palliation was achieved by surgical septation in two of our patients with double inlet and double outlet from the main chamber. However, heart block was surgically induced in both. One patient died postoperatively, and the other died later due to pacemaker failure. Palliation in hearts such as these is theoretically feasible, and a knowledge of the course of the conduction system is therefore vitally significant to the surgeon. In the four hearts we studied histologically (Wilkinson JL, Dickinson DF, Smith A, Anderson RH: submitted for publication), the atrioventricular bundle always arose from a normally situated atrioventricular node and descended onto either the trabecular septum at the crux cordis (three cases) or a posterior trabecula at the crux (one case). This is in marked contrast to univentricular hearts of either left ventricular type or indeterminate type, where histological and electrophysiological studies have shown the connecting atrioventricular conduction system to be located in either the anterior or anterolateral quadrants of the right atrioventricular orifice (fig. 7). Thus, during repair of a univentricular heart of right ventricular type, as with a "huge ventricular septal defect," the crux of the heart is likely to be the danger area. The preoperative diagnosis of a univentricular heart of the right ventricular type therefore indicates the probable position of the atrioventricular bundle, and facilitates the necessary intraoperative mapping procedures which should be carried out during repair.

Nosological Considerations

Many observers, notably Van Praagh et al., have pointed to the apparent paradox of using the term single ventricle to describe a heart with two chambers in its ventricular mass. Nonetheless, the term is still widely used to describe such anomalies. To justify this usage, we propose that one of these chambers cannot be a ventricle.

In single ventricle with outlet chamber, Van Praagh et al. and Lev and his colleagues suggest that the outlet chamber is merely the infundibulum of the right ventricle. Our observations, in contrast, show that the outlet chamber has both an infundibulum and a trabecular zone of right ventricular type, but lacks an inlet portion, defined as the part of a ventricle related to and containing an atrioventricular valve. This distinction is important when considering the hearts we describe, which satisfy all the criteria for inclusion as single or common ventricle, since the rudimentary chambers have the trabecular zone of left ventricular type but lack an inlet portion. We suggest that the criterion for description of a chamber as a ventricle, is that it possesses an inlet portion. Chambers without inlet portions can be defined as rudimentary chambers. However, it is known that ventricles do not always possess outlet portions (for example, the left ventricle in the presence of double outlet right ventricle). Our observations show that rudimentary chambers, similarly, do not always have outlet portions; many of the chambers have only a trabecular
zone. These chambers cannot be outlet chambers, but are still rudimentary. We suggest the term “trabecular pouch” for their distinction, and have observed such chambers not only in the hearts we describe, but with right ventricular trabecular pattern in univentricular hearts of left ventricular type.\(^{13}\)

The present findings are contrary to our previously published views concerning "primitive ventricle." In these papers\(^ {14, 16}\) we argued that all univentricular hearts could be designated as primitive ventricle, defined as absence of the posterior part of the interventricular septum running to the crux cordis. The present cases are unequivocally univentricular but are not primitive ventricles, since they possess a septum which extends to the crux cordis.

They also differ in several other ways. In primitive ventricle with outlet chamber,\(^ {15}\) the main chamber has left ventricular characteristics and the rudimentary chamber has right ventricular characteristics. In univentricular heart of right ventricular type, the main chamber has right characteristics and the rudimentary chamber has left characteristics. In primitive ventricle the rudimentary chamber, when present, is almost always anterior to the main chamber, and in right ventricular type univentricular heart it is always (in our experience) posterior. The conducting tissue disposition is markedly different in the two types of univentricular heart. We therefore believe it is important to distinguish our cases from primitive ventricles.

Our previous arguments regarding primitive ventricle were concerned with establishing that the main chamber of these hearts was not a normal left ventricle which simply had double inlet.\(^ {16}\) Similarly, our present findings indicate that univentricular heart of right ventricular type is not simply a double inlet right ventricle. Therefore, we believe it is more accurate to use the descriptive terms univentricular heart of left ventricular type instead of primitive ventricle with outlet chamber or double inlet left ventricle and univentricular heart of right ventricular type instead of double inlet right ventricle or single right ventricle. The third type of univentricular heart which we previously described as primitive ventricle without rudimentary chamber would be described more accurately as univentricular heart of indeterminate type. We believe that the loss in brevity is compensated for by improved clarity of description.

Hearts with straddling valves are a further problem, since in these hearts a ventricular inlet portion is connected to both chambers in the ventricular mass. Where the degree of straddling is minimal, problems in definition are unlikely since the straddling valve can be treated as if it belonged to the chamber receiving its greater part as in the cases of this series. Cases may, however, arise where the straddling valve is more equally committed to both chambers. In this situation the solution should be pragmatic, assigning the valve to the chamber which appears to receive more than half its circumference. This system has been used to deal with overriding arterial valves.\(^ {16}\)

In summary, the cases described demand an extension of the accepted views concerning single ventricle with outlet chamber\(^ {1}\) since analogous hearts can exist in which the main chamber is of right ventricular type and the rudimentary chamber of left ventricular type. These hearts can be distinguished by noting not only the trabecular characteristics, but also the position of the rudimentary chambers and the orientation of the

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**Figure 7.** Diagrams showing the anticipated disposition of the conducting tissue in univentricular hearts with an anterior trabecular septum running to the acute margin (left ventricular or indeterminate types) compared with those with a trabecular septum running to the crux (right ventricular type).
septal structures. Their categorization is facilitated by the usage of purely descriptive terms.

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Univentricular heart of right ventricular type with double or common inlet.
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