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Original Articles

Cross-Sectional Echocardiographic Measurements of Right Ventricular Size and Growth in Patients with Pulmonary Atresia and Intact Ventricular Septum

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SUMMARY. **Fifteen patients with pulmonary atresia or critical pulmonary stenosis and intact ventricular septum were studied. All were operated on in the neonatal period, with valvotomy or a systemic to pulmonary arterial shunt, or both. In 12 patients, right ventricular to pulmonary arterial communication was established in the neonatal period. In three patients, only systemic to pulmonary arterial shunts were constructed. Six patients died. The median follow-up period for the surviving patients was 64 months (range, 12–87 months).**

Right and left atrial and ventricular dimensions and areas, the tricuspid annular diameter, and the cross-sectional area of the aortic root were measured in cross-sectional echocardiograms from the neonatal period, at the age of 1 year, and at the latest clinical follow-up. A classification of right ventricular morphology was made, based on identification of the inlet, the trabecular, and the outlet parts.

Most of the patients had hypoplastic right ventricles at birth but at the latest follow-up, seven of nine surviving patients had right ventricles in the normal range. Right ventricular growth was better in patients who were given a right ventricular to pulmonary arterial communication in the neonatal period and those with complete right ventricular anatomy. The patients who died had severely hypoplastic right ventricles and small tricuspid valves.

KEY WORDS: **Pulmonary atresia — Cross-sectional echocardiography — Right ventricular size — Right ventricular growth**

In children with pulmonary atresia or critical pulmonary stenosis and intact ventricular septum, the right ventricle may be of normal size or enlarged, but right ventricular hypoplasia, varying from moderate to severe, is more common [5, 6, 15–17]. A classification based on right ventricular size and morphology has been proposed [1, 3]. It has been shown that the creation of continuity between the right ventricle and the pulmonary artery is important for right ventricular growth [1–3, 11, 14, 17]. Only in a few studies [13, 19, 20], however, has cross-sectional echocardiography been used for serial evaluation of these patients.

The aim of our study was to study right ventricular size and growth by cross-sectional echocardiography in patients with severe right ventricular outflow obstruction and intact ventricular septum.

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Material and Methods

Fifteen consecutive patients (seven girls, eight boys) with severe right ventricular outflow obstruction and intact ventricular septum, born between November 1980 and August 1987 and requiring operation during the first week, were studied. Three patients had critical pulmonary stenosis, 10 had pulmonary valve atresia, and two had atresia of the main pulmonary artery. Only patients requiring surgery in the first week of life were included. One child was born prematurely after 33 weeks of gestation; the other patients were born at term. The median birth weight was 3.4 kg (range, 1.5–4.5 kg). All patients presented with cyanosis in the first days of life, and in all cases the diagnosis was established by cross-sectional echocardiography and cardiac catheterization. The median age at operation was 2 days (range, 1–7 days). Twelve patients were treated with prostaglandin before surgery.

In 12 patients, right ventricular to pulmonary artery communication was established, either by valvotomy or by reconstruction of the right ventricular outflow tract with a right ventricular outflow patch. Ten of these patients also had systemic to pulmonary artery shunts. Three patients were initially given only systemic to pulmonary artery shunts.

Table 1. Clinical data of the patients

Patient surgery no.	Age	Age at latest echo (months)	Outcome	Morphological class
Critical pulmonary stenosis				
1. Pulmonary valve extirpation	1 day			I
Blalock-Taussig shunt	1 week			
Modified Blalock-Taussig shunt, atrial septectomy	9 months			
Modified Glenn and Fontan procedure	57 months	75	Alive, well	
2. Valvotomy	2 days			I
Valvotomy, closed ASD	29 months	72	Alive, well	
3. Modified Blalock-Taussig shunt	7 days			II
Right ventricular outflow patch, ligature of sinusoids	9 months	12	Alive, well	
Pulmonary valve atresia				
4. Valvotomy, AP shunt	1 day			I
Closed AP shunt, Waterston, right ventricular outflow patch	3 weeks			
Closed Waterston, pulmonary patch, closed ASD, tricuspid plastic	39 months	87	Alive, well	
5. Valvotomy, AP shunt, right ventricular outflow patch	1 day			II
Closed ASD	33 months	78	Alive, well	
6. Valvotomy	1 day	64	Alive, well	I
7. Valvotomy, AP shunt	1 day		Died at operation	I
8. Right ventricular outflow patch, AP shunt	1 day		Died postoperatively	II
9. Valvotomy, AP shunt	2 days			I
Valvotomy, right ventricular outflow patch, ligature of sinusoid	7 months			
Valvotomy	12 months			
Modified Blalock-Taussig shunt	18 months	42	Alive, well	
10. Valvotomy, AP shunt	2 days			II
Valvotomy, modified Blalock-Taussig anastomosis, ligature of sinusoid	7 months	24	Alive, well	
11. Right ventricular outflow patch, AP shunt	2 days			II
Modified Blalock-Taussig shunt	18 months	19	Died 33 months	
12. Valvotomy, AP shunt	1 day			I
Resection infundibular stenosis, right ventricular outflow patch, modified Blalock-Taussig shunt	10 months			
Biological pulmonary valve prosthesis	21 months	21	Alive, well	
13. Valvotomy, AP shunt	2 days		Died operatively	I
Pulmonary artery atresia				
14. AP shunt	5 days		Died postoperatively	I
15. Modified Blalock-Taussig shunt	2 days	4	Died at 6 months	III

AP, arteriopulmonary.

The patients' diagnosis, morphological classification of the right ventricles, initial and later surgical procedures, clinical outcome, and age at the latest echocardiographic examination are listed in Table 1. There was angiographic evidence of myocardial sinusoids in patients 3, 8, 9, 10, 11, 14, and 15. These patients all had severely hypoplastic right ventricles at birth. In patient 15 there was angiographic evidence of a small, hemodynamically insignificant defect in the muscular part of the interventricular septum.

Four patients died early, either during operation or in the immediate postoperative period. For the rest of the children the median follow-up period was 42 months (range, 4–87 months). In this group, another two children died (patients 11 and 15), at the age of 33 and 6 months, respectively. At autopsy the systemic to pulmonary arterial shunts were open in both cases. As no other obvious cause of death was found, these two children presumably died from ventricular arrhythmia. All surviving patients but one (patient 6) have had additional operations after the neonatal period (Table 1).

In all cases, serial cross-sectional echocardiographic results were available. The first neonatal examination, one study at the age of approximately 1 year, and the latest cross-sectional echocardiogram were used for the measurements. For technical reasons the first examination in patient 4 could not be analyzed. In this patient an examination at the age of 3 weeks was used for the first measurements. In the remainder of the patients the first echocardiogram, at the median age of 1 day (range, 0.5–4 days), was analyzed. The 1-year follow-up study includes 10 patients,

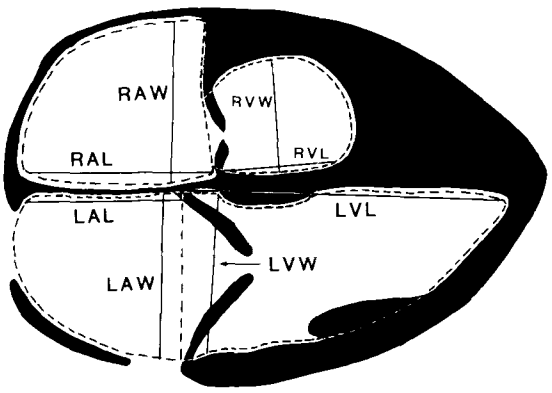


Fig. 1. Measurements in the apical and subcostal four-chamber view. *A*, area; *L*, length; *LA*, left atrium; *LV*, left ventricle; *RA*, right atrium; *RV*, right ventricle; *W*, width; right and left atrial and ventricular areas are marked by *broken lines*.

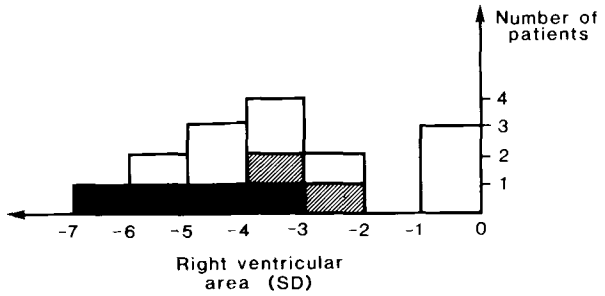


Fig. 2. Right ventricular area in the neonatal period, related to survival or age at death: □, alive; ▨, dead after the first week; ■, dead in the first week.

examined at the median age of 12 months (range, 9–15 months). The median age at the latest examination was 52 months (range, 18–87 months). Echocardiography had been performed with one of three different mechanical sector scanners (ATL Mark 100, Diasonics CV 400, and Vingmed CFM 700) with transducers operating at 5.0 and 7.5 MHz, and recorded on videotape. A computerized system (Cardio 80) consisting of a computer, a digitizing table, a digitizing pen, and a videomonitor connected to a videorecorder, was used for measurements. The technique used for this, as well as the normal values (based on body surface area) have been published previously [10]. Left and right atrial and ventricular length, width, and area were measured in apical or subcostal four-chamber views, as illustrated in Fig. 1. The measurements were taken in the views giving the largest atria and ventricles. The dimension of the pulmonary artery at the level of the pulmonary valve and the cross-sectional area of the aortic root were measured in the parasternal short-axis view. In addition, the diameter of the tricuspid annulus was measured in the apical or the subcostal four-chamber view, as described by Gutgesell et al. and King et al. [9, 12]. This diameter was defined as the largest clearly identifiable distance between the attachments of the tricuspid leaflets, in the four-chamber view.

The angiocardiograms from the neonatal period were also reviewed and classified into three groups according to Bull et al. [1]. In group 1, all three portions of the right ventricle (the inlet, the trabecular, and the outlet parts) were present. In group 2, the trabecular part was missing, and in group 3 both the trabecular and the outlet parts were missing. For technical reasons the angiocardiogram in patient 12 could not be used for the morphological classification. In this patient, therefore, the morphological classification was based on studies of the cross-sectional echocardiogram from the neonatal period. The results of the morphological classification are presented in Table 1. The angiocardiograms were also reviewed for signs of tricuspid regurgitation, classified as significant or absent. In five patients (5, 6, 7, 8 and 10), the presence or absence of tricuspid regurgitation could not be assessed angiographically due to the inadequate position of the catheter.

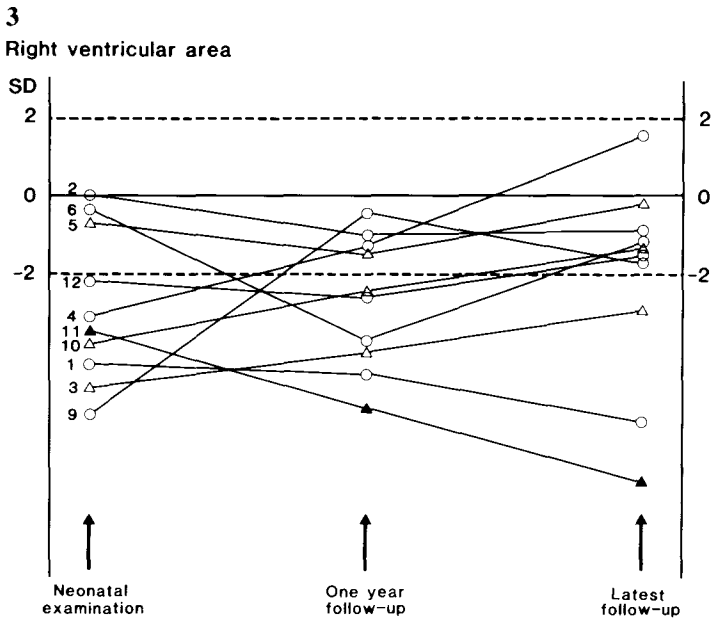
To make the measurements comparable, irrespective of the age and the size of the children, all measurements—except the tricuspid annulus diameter—were expressed in standard deviations (SD) from the normal mean, based on body surface area [10]. The normal range for a single patient was ±2 SD. However, for a group of normal patients the normal mean value is 0 SD. When measurements were available in both the apical and the subcostal four-chamber views, the value closest to normal was used for further analysis. The diameter of the tricuspid annulus is presented in millimeters.

The Student's *t*-test was used for comparisons between the group mean values and the normal mean values and for comparisons between different groups in the material. Statistical significance was defined as $p < 0.05$ for the difference between the compared mean values.

Results

Thirty-five cross-sectional echocardiograms, 15, from the neonatal period, 10 from the 1-year study, and 10 from the latest study, were analyzed. In the neonatal period, 75% of the analyzed four-chamber echocardiograms were subcostal recordings. At the age of 1 year and later, about 50% of the four-chamber echocardiograms used for the analysis were apical and 50% were subcostal. The results of measurements of length and width were similar but less informative than the corresponding area measurements and are therefore not presented. No significant differences were found between the group of patients with critical pulmonary stenosis and that with pulmonary atresia. Therefore, the results are presented together.

Right ventricular area in the neonatal period, related to clinical outcome, is presented in Fig. 2. Most of the patients had clearly hypoplastic right ventricles initially. The mean area of the right ventricle in the neonatal period in the patients who died was −4.4 SD, compared to −2.8 SD for the surviving patients, but the difference was not statistically significant. Right ventricular size and growth in the 10 children who survived the first year of life is shown in Fig. 3 where the results of the morphological classification are also indicated. As can be seen,



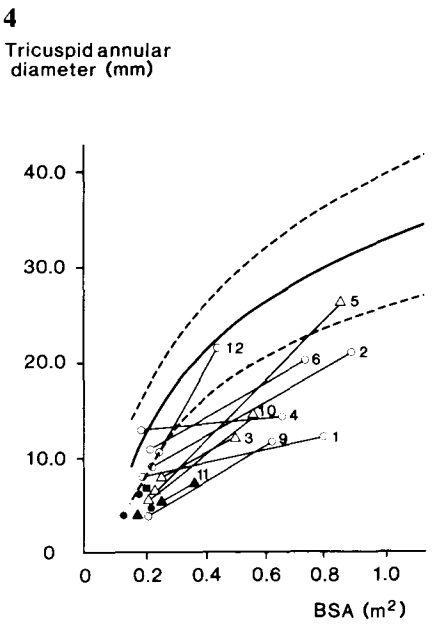


Fig. 3. Right ventricular growth in the children who survived the first year of life. The measurements are given as standard deviations from the expected normal values for each child (numbered as in Table 1). The patient who died is indicated by a black symbol. ○, complete right ventricular morphology; △, right ventricle lacking the trabecular part; and □, right ventricle lacking the trabecular and outlet parts.

Fig. 4. Diameter of the tricuspid annulus in the neonatal period and at the latest follow-up. The normal range is that given by King et al. [12]. Symbols as given in Fig. 3. The patients who died are indicated by black symbols.

seven patients had a right ventricular area within normal range at the latest follow-up. Three of these patients had clearly hypoplastic right ventricles at birth.

Measurements of tricuspid valve annulus diameter in the neonatal period and at the latest follow-up are shown in Fig. 4 and compared with the normal values established by King et al. [12]. Almost all of the patients had somewhat hypoplastic tricuspid valves. A smaller tricuspid annulus was found in the patients with a smaller right ventricle. The patients with incomplete right ventricular anatomy (groups 2 and 3) had tricuspid annular diameters below the normal range for body surface area at birth. Significant tricuspid regurgitation was found in patients 4, 9, 12, 13, and 14. No statistically significant differences regarding right atrial, right ventricular, or tricuspid annular size or growth were found between the groups with and without tricuspid regurgitation.

The relationship between the right ventricle area and the diameter of the tricuspid valve at birth and the morphological classification are shown in Fig. 5. As can be seen, some patients with complete right ventricular morphology had clearly hypoplastic right ventricles and tricuspid valves, while one patient with incomplete right ventricular morphology had a right ventricle in the normal range.

The results of right atrial area measurements in the patients who survived the first year of life are presented in Fig. 6. All group mean values of right atrial area measurements were significantly larger than normal. In the neonatal period the group mean value of right atrial area in patients with tricuspid regurgitation was +1.8 SD compared to +0.8 SD for patients without tricuspid regurgitation, but the difference was not statistically significant.

Measurements of the diameter of the pulmonary artery at the valve level are presented in Fig. 7. Pulmonary artery diameter was not measured in the patients with atresia of the main pulmonary artery (patients 14 and 15). In patients 1, 4, 11, and 12, one or two (patients 1 and 11) of the three studied examinations were unsatisfactory for measurement. The mean value of the pulmonary artery diameter was −1.4 SD for patients with pulmonary stenosis and −4.2 SD for patients with pulmonary valve atresia. The difference was not statistically significant, but the two groups were small.

The mean and range of the measurements of the left atrium, the left ventricle, and the aortic root are presented in Table 2. The group mean values of the measurements of the left atrium and the aortic root in the neonatal period and at the 1-year follow-up were significantly larger than normal. At the latest

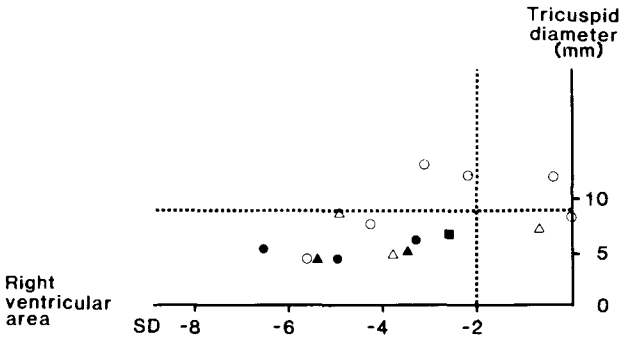


Fig. 5. Relationship between right ventricular morphology, right ventricular size, and tricuspid annular diameter in the neonatal period. The *dotted lines* indicate the lower limits of the normal range for measurements of the tricuspid valve and the right ventricle. As the measurements of the tricuspid valve are given in millimeters, the lower normal limit (8 mm) indicated in the figure is approximated for a newborn child with a body surface area of 0.20 m^2. Symbols as given in Fig. 3. The patients who died are indicated by black symbols.

follow-up the mean values were not significantly different from normal, but the variation among the patients was great, and in some of the patients the left atrium and the aortic root were still clearly enlarged. No significant differences from normal were found with regard to measurements of the left ventricle.

Discussion

Different angiographic [14, 17, 18] and echocardiographic [13, 20] methods have been used to assess right ventricular size in patients with pulmonary atresia or critical pulmonary stenosis and intact ventricular septum. As the shape of the hypoplastic right ventricle is different from normal, the general assumptions of geometry and their application in calculating right ventricular volume are less valid.

The results of our study of the size of the right ventricle, based on cross-sectional echocardiographic measurements, mainly agree with earlier ones in which the right ventricle was studied angiographically or at postmortem [1, 2, 8, 11, 17, 21]. We have found a range from normal to severely hypoplastic right ventricles. As can be seen in Fig. 2, all patients who died had hypoplastic right ventricles at birth, but there was an overlap with the group of surviving patients, and no statistically significant differences were found between those surviving and those who died. At the latest follow-up, seven of nine surviving patients had right ventricles within the normal range. Three of these (patients 4, 9, and 10) had clearly hypoplastic right ventricles at

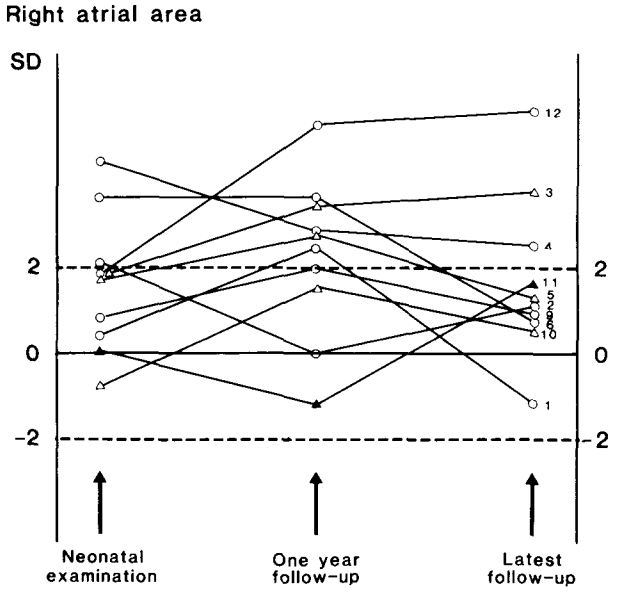


Fig. 6. Right atrial area in the children who survived the first year of life. The values are given in SDs from the expected normal values. Symbols as given in Fig. 3.

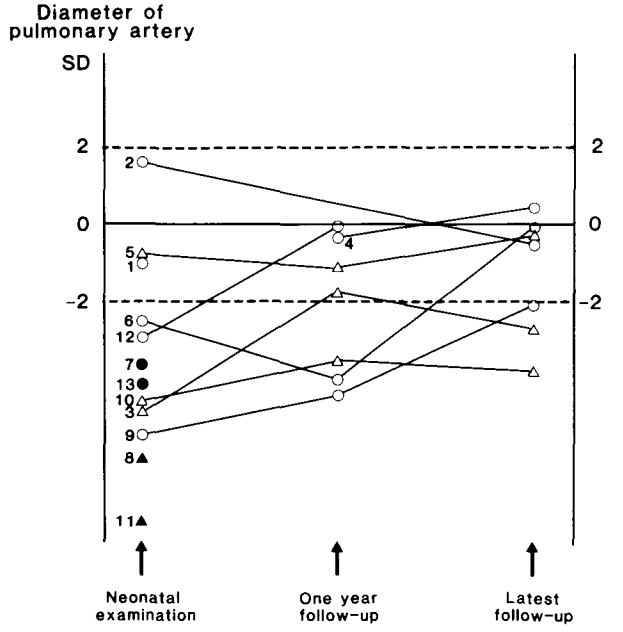


Fig. 7. Diameter of the pulmonary artery at the valve level in the patients with pulmonary stenosis or pulmonary valve atresia. The values are given as SDs from the expected normal values for each child. Symbols as given in Fig. 3. The patients who died are indicated by black symbols.

birth. In all patients with a normal right ventricle at follow-up, right ventricular to pulmonary artery communication had been established in the neonatal period.

Table 2. Measurements of the left atrium, left ventricle, and aortic root

	Neonatal examination	1-year follow-up	Latest follow-up
Left atrium	+1.0 (−1.2–+4.5)[a]	+1.9 (−0.7–+5.0)[a]	+1.5 (−2.3–+7.2)
Left ventricle	−0.6 (−3.5–+2.7)	+0.4 (−3.2–+2.8)	−0.3 (−3.2–+4.4)
Aortic root	+1.2 (−1.6–+3.1)[a]	+2.1 (−3.0–+4.7)[a]	+1.5 (−0.9–+4.8)

Values are expressed as SD. The results are given as the group mean and range (parentheses) for the measurements.

[a] Significantly different ($p < 0.05$) from the normal group mean value (0 SD).

In three patients (patients 1, 3, and 11) right ventricular growth into the normal range was not achieved. In patients 1 and 11, right ventricular to pulmonary artery communication was created in the neonatal period. Postoperative catheterization at the age of 8 months in patient 11, however, showed no open communication from the right ventricle to the pulmonary artery. Patient 3 was operated on at the age of 1 week with a systemic to pulmonary arterial shunt. Thus, two of three patients with inadequate growth of the right ventricle had absent or ineffective flow through the right ventricle during their first months of life. Our findings support the results of several others [2, 4, 6, 11, 14, 16, 17] and show that early creation of a right ventricular to pulmonary arterial communication is important for right ventricular growth.

The dimension of the tricuspid valve has been shown to correlate with right ventricular size in patients with critical right ventricular outflow obstruction [1–3, 10, 16, 17, 20]. Measurements of tricuspid valve annular diameter by cross-sectional echocardiography correlate well with postmortem findings [9], and normal echocardiographic values for tricuspid valve annular diameter have been established [12]. We have found a small tricuspid annulus in almost all of the studied patients and, as could be expected, the smallest tricuspid valves were found in those with extremely hypoplatic right ventricles. In cases with pulmonary atresia and intact ventricular septum, varying abnormalities of the tricuspid valve are frequently found [1, 5, 7, 21]. In our material no patient had Ebstein's anomaly.

The size of the tricuspid valve and the right ventricle, as well as right ventricular morphology, are factors that have been studied in order to predict the prognosis and estimate the value of different surgical procedures in these patients [1, 3, 11, 13, 17, 18]. Tricuspid and right ventricular size are closely related and it has been shown that the combination of a restrictive tricuspid valve and severe right ventricular hypoplasia negatively affects the prognosis [20]. But our data show that some extremely small right ventricles with small tricuspid valves may grow after pulmonary valvulotomy.

A morphological classification based on the identification of the inlet, trabecular, and outlet parts of the right ventricle has been proposed by Bull et al. [1]. Several workers [2–4, 14] have shown that only right ventricles with an identifiable outlet part grow after valvotomy, whereas the absence of the trabecular part does not preclude right ventricular growth. In two of our patients (5 and 10), with normal right ventricular size at the latest follow-up, the outlet parts were present but extremely narrow at birth. Only one patient lacking both the trabecular and the outlet parts of the right ventricle (patient 15) was included in our study. He was given a systemic to pulmonary arterial shunt and died at the age of 6 months. At birth, his right ventricular area was −2.6 SD and at the last examination, at the age of 4 months, it was −1.6 SD: some growth had taken place. In contrast to Bull et al. [1] we found no clear correlation between tricuspid annular diameter and the morphological classification (Fig. 5). But the three children with tricuspid annular diameter within the normal range all had complete right ventricular morphology.

Although hypoplastic right ventricle, small tricuspid annulus, and incomplete right ventricular morphology were more common in the group of patients who died, we found a clear overlap with the group of surviving patients. Some severely hypoplastic right ventricles had the capacity to grow, and incomplete right ventricular anatomy did not exclude a right ventricle of normal size (patient 5) or right ventricular growth into the normal range following valvotomy (patient 10).

It has been shown that myocardial sinusoidal coronary artery communications are common in the hypertensive and severely hypoplastic right ventricles [5, 6, 11]. All of our patients with angiographic evidence of myocardial sinusoids had severely hypoplastic right ventricles at birth. Myocardial sinusoids were found in four of six patients who died, including the two late deaths.

We found significant enlargement of the right atrium for the group as a whole. Figure 6 indicates an increase in right atrial area during the first year with a decrease in most patients in later examinations.

As in earlier studies [13, 21], we found no correlation between the size of the right ventricle and the dimension of the pulmonary artery.

Miller et al. [15] showed that the diameter of the pulmonary artery was larger in patients with critical pulmonary stenosis than in those with pulmonary atresia. In our material the difference in pulmonary artery diameter between the two groups was not significant. Bull et al. and Cobanoglu et al. [1, 2] found that patients with critical pulmonary stenosis and intact ventricular septum had less restrictive tricuspid valves and tended to have larger right ventricles than those with pulmonary atresia and intact ventricular septum. These results were not confirmed in our study as two of our three patients with critical pulmonary stenosis had severely hypoplastic right ventricles with have not grown adequately. However only three patients with pulmonary stenosis are included, and all of these required surgery in the first week of life. Different results may be obtained if patients with critical pulmonary stenosis requiring surgery later than the first week were included, as in the studies by Bull et al., Cobanoglu et al. and Miller et al. [1, 2, 15].

To assess the degree of right ventricular hypoplasia, the ratio between left- and right-sided measurements has been used [8, 14]. Fricker et al. [8] calculated the ratio between right and left ventricular inlet and outlet dimensions, whereas Lewis et al. [14] used the ratio between the diameter of the descending aorta and a right ventricular index, based on the measurement of the tricuspid valve and the inlet and outlet portions of the right ventricle.

We found enlargement of the left atrium and the ascending aorta, reflecting the increased blood flow through the left side via the foramen ovale or an atrial septal defect. If left-sided valves are included in the assessment of right ventricular size, only measurements that are not affected by right-to-left shunting or volume load of the left ventricle should be used for calculating the ratios between right- and left-sided measurements.

Measurements in cross-sectional echocardiography have their limitations [10] and considerable differences are necessary to be sure that a real change has taken place. The abnormal shape of the right ventricles in patients with pulmonary atresia and intact ventricular septum increases the difficulties of identifying the proper anatomical landmarks used for measurement. These limitations, however, also affect angiographic methods of determining right ventricular size in these patients.

Despite the limitations our study indicates that cross-sectional echocardiography is a useful method for serial evaluation of right ventricular size and development in patients with severe right ventricular outflow obstruction. We have found right ventricular size ranging from normal to severely hypoplastic. The prognosis is worse for patients with very small right ventricles, but even severely hypoplastic right ventricles and right ventricles with incomplete morphology may grow. Early creation of a communication between the right ventricle and the pulmonary artery is important for stimulating growth of the right ventricle.

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