Angiographic Study of Univentricular Heart of Right Ventricular Type

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SUMMARY An angiographic analysis of 10 cases of univentricular heart of the right ventricular type is reported. This congenital malformation is characterized by a large chamber with right ventricular morphology that receives both atrioventricular valves, and a second, smaller chamber, a trabecular pouch, with left ventricular morphology. These chambers are separated by a posterior septum but are connected by an inlet septal defect.

The angiographic studies were done using the angled angiographic techniques in three patients and the standard frontal and lateral angiographic views in seven cases. The atrial situs in seven patients was solitus, in one inversus, and in two it was ambiguous with left isomerism. In seven patients the usually large right ventricular chamber received two atrioventricular valves and in four patients, one atrioventricular valve was straddling. Three patients had atresia of one atrioventricular valve.

The trabecular pouch was small in seven patients but relatively large in three. In six patients the trabecular pouch was located posterior and to the left of the right ventricular chamber and in four anterior and to the right. Double outlet right ventricle was present in all cases. The aorta arose anteriorly to the pulmonary artery in nine patients and posteriorly in one.

An autopsy was performed in one case and its correlation with the angiographic findings was remarkable. The angiographic demonstration of the anatomical details of this entity and its associated anomalies was facilitated by angled angiography.

Univentricular Heart is a congenital malformation in which a single ventricular chamber receives the entire atrial outputs, through two atrioventricular valves or a common valve when one valve is absent. Three types of univentricular heart can be identified, depending on the anatomic features of the ventricular chamber receiving the valve or valves: left ventricular type, right ventricular type, and indeterminate type.

Among univentricular hearts, the left ventricular type is the most frequent, the easiest to diagnose angiographically, and from the surgical viewpoint, is most amenable to total correction. The right ventricular type is a rare condition, and although some angiographic studies have been reported, they do not clearly define the anatomy. In this report we analyze the angiographic anatomy of 10 cases of univentricular hearts of right ventricular type seen at the University of Alabama in Birmingham over 8 years.

Definition of Terms

Ventricular Morphology

Univentricular heart of right ventricular type is defined as a ventricular malformation characterized
by a large chamber with the trabeculated pattern of the right ventricle receiving two atrioventricular valves or, in the absence of one valve, the sole atrioventricular valve. Such hearts usually have a septal structure that extends toward the crux of the heart. A rudimentary chamber with the trabecular pattern of the left ventricle is present, but usually does not have either inlet or outlet portions. This chamber may be called a trabecular pouch. In rare circumstances the rudimentary chamber may have an outlet and is then termed an outlet chamber. We did not see such hearts in this study, but they have been described. Cases without rudimentary chamber were also excluded, since they cannot be differentiated from the indeterminate type with certainty. Cases with absence or atresia of one atrioventricular valve are included in this study only when an anatomic potential communication between the blind-ending atrium and the large ventricular chamber is demonstrated, or else when it was clear that the blind-ending atrium had no possible potential connection with the rudimentary chamber.

We use the terms dextrocardia and levocardia to indicate the position of the cardiac axis independent of the connections or relation of the heart chambers and great vessels. The relationship between the main and rudimentary chamber is described in terms of anteroposterior, superoinferior and right-left positions.

The atrioventricular valves are called right, left or common, according to their anatomic structure and their connection with the corresponding atrium. We avoid the terms mitral and tricuspid because the morphology of the atrioventricular valves in univentricular hearts is usually not similar to that of the normal valves.

Straddling atrioventricular valve is a condition in which the right or left atrioventricular valve overrides the septum to some degree. We consider the straddling atrioventricular valve a transitional stage between the biventricular hearts and double inlet. We assign the straddling atrioventricular valve to the chamber underlying more than 50% of such valve.

The arterial connection establishes the anatomic relationship between the ventricular chambers and the great arteries. In this report, since by definition the arteries are related only to the main chamber, the connection is either double outlet or single outlet. In cases in which only one great vessel arises from the heart, we use the terms single outlet of the heart with pulmonary atresia or aortic atresia.

**Material and Methods**

Ten patients who had a main ventricular chamber with the morphological appearance of right ventricle and a trabeculated pouch with left ventricular morphology are the subjects of this study. This diagnosis was made angiographically. One patient also had an autopsy study.

Catheterization was performed with the patients under general anesthesia. NIH catheters were introduced from the saphenous vein in seven patients and from the axillary vein in three.

The angiographic study was made by selective venous catheterography of the main chamber. Selective opacification of the trabecular pouch was performed in three patients. Additional selective injections of contrast media were made beneath the semilunar valves in three patients in whom the ventriculograms failed to demonstrate the anatomy of this area. Renografin-76 (meglumine diatrizoate) was injected by hand in small children and by power injector in larger patients. In patients younger than 1 year, 1 ml of contrast media per pound per injection was used. Each injection was separated by a 15-minute interval (the estimated time for clearance of 40% of the contrast from the vascular system). In larger pediatric
patients and adults, 40–60 ml of contrast media per injection was delivered at 30 ml/sec.

Frontal and lateral views were used for cineangiograms in seven cases. In three cases, biplane axial angiography was used. The basic axial projections were the “four-chamber” and elongated right anterior oblique views. In our series two of three patients studied with axial angiograms needed three injections of contrast media, and two injections were done in the third patient.

Angiographic Study

In the angiographic analysis of the univentricular heart, a specific protocol has been followed: 1) identification of atrial situs and its venous connections; 2) analysis of the number and mode of connection of the atrioventricular valves; 3) analysis of the ventricular anatomy including the rudimentary chamber; 4) analysis of the ventriculoarterial connections; and 5) associated anomalies. In each of these steps, the following angiographic features were considered for the final analysis:

Identification of the Atrial Situs and Its Connections

The angiographic features leading to the diagnosis of the atrial situs have been previously reported.

Angiographic demonstration of atrioventricular connections was made using procedures previously reported.

Analysis of the Number and Mode of Connections of the Atrioventricular Valves

In the four-chamber view, the angiographic anatomy of the posterior segment of the ventricular septum is usually best visualized and overriding of the atrioventricular valves identified. A double inlet connection is established when two atrioventricular valves were seen to connect the greater part of both atria with the same ventricular chamber. An absent atrioventricular connection (single inlet) is established when one atrium has no direct communication with the rudimentary ventricular chamber. It may have had a potential connection with the main chamber. The right atrioventricular valve is usually higher and more

**Figure 2.** Diagram of the atrioventricular (AV) connection mode in univentricular heart, right ventricular type. This arrangement may be also in situs inversus or ambiguous.

**Figure 3.** Right ventriculogram in lateral view (case 6). The right ventricular chamber (RV) is very large and is divided into three segments by two filling defects (arrowheads) located in the anterior wall, which may represent short muscular ridges or papillary muscles. The right and left atrioventricular valves (RAV and LAV) enter completely the right ventricular chamber. The small trabecular pouch that is not seen in this angiogram is located posteriorly and to the left of the main chamber. The great arteries arise from the anterior portions of the main chamber in a side-by-side relationship. AO = aorta; PA = pulmonary artery.
Table 1. **Angiographic Features of Univentricular Heart of Right Ventricular Type**

<table>
<thead>
<tr>
<th>Cases</th>
<th>Age</th>
<th>Sex</th>
<th>Heart axis</th>
<th>Situs</th>
<th>AV Valves (number)</th>
<th>Mode of AV connection</th>
<th>Main chamber angiographic anatomy</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>22 mos</td>
<td>M</td>
<td>Right</td>
<td>Inversus</td>
<td>Single</td>
<td>Atresia of the right</td>
<td>Mainly LV pattern (RV pattern in anterior wall)</td>
</tr>
<tr>
<td>2</td>
<td>6 yrs</td>
<td>M</td>
<td>Left</td>
<td>Solitus</td>
<td>Double</td>
<td>Straddling on the left</td>
<td>Mainly RV pattern (LV pattern in posterior wall)</td>
</tr>
<tr>
<td>3</td>
<td>29 mos</td>
<td>M</td>
<td>Left</td>
<td>Solitus</td>
<td>Single</td>
<td>Atresia of the left</td>
<td>Mainly RV pattern (LV pattern in posterior wall)</td>
</tr>
<tr>
<td>4</td>
<td>27 yrs</td>
<td>F</td>
<td>Left</td>
<td>Ambiguous left</td>
<td>Double</td>
<td>Straddling of the right</td>
<td>RV pattern</td>
</tr>
<tr>
<td>5</td>
<td>1 yr</td>
<td>M</td>
<td>Left</td>
<td>Solitus</td>
<td>Double</td>
<td>Straddling of the right</td>
<td>Mainly RV pattern (LV pattern in posterior wall)</td>
</tr>
<tr>
<td>6</td>
<td>10 yrs</td>
<td>M</td>
<td>Left</td>
<td>Solitus</td>
<td>Double</td>
<td>Complete connection with RV</td>
<td>Mainly RV pattern (LV pattern in septal wall)</td>
</tr>
<tr>
<td>7</td>
<td>3 yrs</td>
<td>F</td>
<td>Right</td>
<td>Solitus</td>
<td>Double</td>
<td>Complete connection with RV</td>
<td>Right ventricular pattern</td>
</tr>
<tr>
<td>8</td>
<td>4 yrs</td>
<td>F</td>
<td>Left</td>
<td>Solitus</td>
<td>Double</td>
<td>Straddling of the left</td>
<td>Right ventricular pattern</td>
</tr>
<tr>
<td>9</td>
<td>11.5 yrs</td>
<td>M</td>
<td>Left</td>
<td>Solitus</td>
<td>Double</td>
<td>Complete connection with RV</td>
<td>Mainly RV pattern (LV pattern in posterior wall)</td>
</tr>
<tr>
<td>10</td>
<td>5 yrs</td>
<td>F</td>
<td>Right</td>
<td>Ambiguous left</td>
<td>Single</td>
<td>Atresia of the left</td>
<td>Mainly RV pattern (LV pattern in posterior wall)</td>
</tr>
</tbody>
</table>

Abbreviations: AV = atrioventricular; PA = pulmonary artery; AVV = atrioventricular valve; AO = aorta; Assoc Anom = associated anomalies; RV = right ventricle; LV = left ventricle; Discont. = discontinuity; Cont. = continuity; VIF = ventricular infundibular fold; PS = pulmonary stenosis; ASD = atrial septal defect; LSVC = left superior vena cava; RVI = right valve insufficiency; ARPA = absence of right pulmonary artery.

In some cases, however, it is in the same sagittal plane. Straddling right or left atrioventricular valves are established when the annulus alone or the annulus and the peripheral apparatus are inserted in both sides of the septum (fig. 1). The angiographic demonstration of leaflets and chordae tendineae is more easily visualized on motion picture than on single frame. The valves can be identified by a negative shadow during diastole that encroaches on the contrast containing the trabecular pouch and the large ventricular chamber. When the atrioventricular connection is made through a single valve, such a valve may consist of a large common atrioventricular valve or a well-developed right valve with absence of the left valve or vice versa (fig. 2). The distinction between a single atrioventricular valve (absence of one atrioventricular connection) and a common atrioventricular valve can be made by cineangiographic study: The common atrioventricular valve is connected with both atria and its two functional leaflets open and close in an “eyelid” fashion perpendicular to the ventricular axis.

**Figure 4.** Right ventriculogram in four-chamber view (A) and right anterior axial oblique view (B). A) Case 9. The right ventricle (RV) is a large chamber receiving the right and left valves (RAV and LAV). Discontinuity between the aortic and atrioventricular valves is seen. The aorta (Ao) obscures the pulmonary artery, which is small. The trabeculated pouch is not seen in this view. B) The RV is seen in profile. The atrioventricular valves are superimposed; the RAV is higher than the LAV. The trabeculated pouch (TP) is seen in profile attached to the inferior part of the right ventricle.
Table 1. (Continued)

<table>
<thead>
<tr>
<th>Trabecular pouch</th>
<th>Ventriculoarterial connection</th>
<th>PA-AVV relationship</th>
<th>AO-AVV relationship</th>
<th>Aortic Arch</th>
<th>Assoc Anom</th>
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</thead>
<tbody>
<tr>
<td>Posterior left</td>
<td>Double outlet</td>
<td>Discont.</td>
<td>Cont.</td>
<td>Left</td>
<td>PS</td>
</tr>
<tr>
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<td>Double outlet</td>
<td>Discont.</td>
<td>Discont.</td>
<td>Left</td>
<td>ASD</td>
</tr>
<tr>
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<td>Double outlet</td>
<td>Discont.</td>
<td>Discont.</td>
<td>Left</td>
<td>PS, LSVC</td>
</tr>
<tr>
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<td>Double outlet</td>
<td>Discont.</td>
<td>Discont.</td>
<td>Right</td>
<td>PS, RVI</td>
</tr>
<tr>
<td>Posterior left</td>
<td>Double outlet</td>
<td>Cont.</td>
<td>Discont.</td>
<td>Left</td>
<td>ASD</td>
</tr>
<tr>
<td>Inferior right</td>
<td>Double outlet</td>
<td>Discont.</td>
<td>Discont.</td>
<td>Left</td>
<td>PS, ASD</td>
</tr>
<tr>
<td>Inferior right</td>
<td>Double outlet</td>
<td>Discont.</td>
<td>Discont.</td>
<td>Left</td>
<td>PS, ASD, ARPA</td>
</tr>
<tr>
<td>Posterior left</td>
<td>Double outlet</td>
<td>Discont. Large VIF</td>
<td>Discont. Large VIF</td>
<td>Left</td>
<td>PS, ASD</td>
</tr>
</tbody>
</table>

Analysis of the Ventricular Anatomy Including the Rudimentary Chamber

The morphology of the main chamber is identified according to the trabecular pattern as either right or left. When the trabeculation pattern is not clearly left or right type, the ventricle is called indeterminate. The main chamber usually is a large, bulbous structure with a rounded apex. The trabeculations are heavy in most cases, but they are not distributed uniformly. Fine trabeculations primarily of the posterior and inferior walls suggest an undifferentiated type of ventricle. However, when a trabecular pouch with left ventricular type trabeculations is present as a separate

Figure 5. Right ventricular angiograms of two patients obtained on lateral (A) in one patient and long-axial (B) views in the other. A) Case 5. The right ventricular chamber (RV) is the largest chamber located anterior to the trabecular pouch (TP). The right atrioventricular valve was not visualized in this angiogram. The left atrioventricular valve (LAV) connects the left atrium with the main chamber. Part of the annulus and leaflets of the LAV are in contact with the TP, indicating a complete form of straddling valve. The great arteries arise from the main chamber, with the aorta (Ao) anterior to the pulmonary artery (PA). A short infundibular septum (IS) separates the subaortic and subpulmonary portions of the right ventricular outflow tract. B) Case 1. The RV is anterior to the TP. The left LAV connects the left atrium with the ventricle. There was absence of the right atrioventricular valve. The TP is a small posterior chamber connected with the ventricle through a small septal defect (sd) that is located near the aortic valve. There is continuity between the LAV and Ao. The great arteries arise from the ventricle in a normal relationship, with the PA anterior and to the left of the Ao.
chamber, it makes the right ventricular domination unequivocal. A group of large trabeculations may be present at the posterior wall of the right ventricle, forming a posterior ridge; this structure may be incorrectly interpreted as a ventricular septum, resulting in an erroneous diagnosis of a heart with three "ventricular" chambers (fig. 3). The trabecular pouch of left ventricular type is seen as a ventricular chamber similar to those of a normal left ventricle with a smooth wall adjacent to and connected with the main ventricle (figs. 1, 4B, 5-7).

Analysis of the Ventriclearterial Connection

This connection is easy to establish angiographically. The great arteries arising from the heart may have several interrelations, as reported by several au-
The position and level of the arterial valves are best seen in the diastolic phase (figs. 5A and 5B). Particular attention must be taken in studying the continuity between the arterial and atrioventricular valves (figs. 7 and 8).

Associated Anomalies

Associated anomalies should be mentioned in order of importance.

Results

A summary of the angiographic features of our 10 cases is shown in table 1. There were six males and four females in the series. The age range was from 22 months to 27 years. Seven cases had a situs solitus of the atria, one had a situs inversus and the other two cases had a situs ambiguous (left isomerism).

Two atrioventricular valves were present in seven cases (figs. 1, 3, 4 and 7), the left atrioventricular valve
Figure 8. Anatomic appearance of the case shown in figure 7 (case 8). A) Anterior view of the main chamber in its right portion. The bivalved right atrioventricular valve (RAV) connects the right atrium (RA) and morphological right ventricular chamber (MRV). The ostium infundibulum (OI) is the connection between the right and left portions of the chamber. There is a muscular septal defect (sd) located posteriorly near the crux of the heart, which connects this chamber with the trabecular pouch (TP) seen in C. A large ventriculoinfundibular fold (VIF) separates the RAV from the aorta (Ao). The infundibular septum (IS) forms the left wall of the subaortic outflow tract. Inlet (B) and outlet (D) segments of the left portion of the ventricle are shown. The left atrioventricular valve (LAV) connects this chamber with the left atrium (LA). A muscular septal defect connects the morphological MRV with the TP as seen in C. The outlet segment of this part of the ventricle is formed by a large VIF posteriorly, by the IS in its right aspect and by the free wall anteriorly. A large papillary muscle supports the anterior leaflet of the LAV. The left aspect of the OI is seen beneath the IS. The pulmonary valve originates entirely from the right ventricle and it is in discontinuity with the LAV. A short segment of this valve straddles the posterior septum (SV). C) Inferior view of the TP. The free wall has been lifted for better exposure. The walls are smooth similar to the normal left ventricle. No atrioventricular valve is seen entering this chamber. The trabecular septum (TS) makes up the superior wall of the TP. Two muscular septal defects were identified. The superior is connected with the right part of the ventricle as seen in A. The inferior septal defect is in connection with the left part of the ventricle as seen in B. There is a third septal defect in the left corner surrounded partially by fibrous tissue of the LAV. Chordae tendineae pass from the ventricle into the TP (arrowhead), indicating a peripheral form of straddling LAV.
was absent in two cases and the right atrioventricular valve was absent in the other case (fig. 5B). Two right and two left atrioventricular valves were straddling the ventricular septum (figs. 1, 5A and 6). The angiographic features of the main chamber were as follows: 1) The inlet portion just beneath the atrioventricular valves was heavily trabeculated and similar to a normal right ventricle in all cases (fig. 4A). 2) The trabeculated portion was, in nine out of 10 cases, similar to a normal right ventricle except at the level of the posterior wall in which a left ventricular pattern was present (fig. 4B). In the tenth case, a left ventricular trabeculation pattern was present throughout most of the main chamber except for a small segment of the anterior wall (fig. 5B). 3) The outlet portion was usually large and gave rise to the aorta and the pulmonary artery in all of our cases (figs. 4 and 5A).

The infundibular septum was identified separating the subarterial segments (figs. 7C and 7D). In one out of 10 cases, the infundibular septum fused shortly after its origin from the anterior wall, giving the appearance of an anterior rudimentary chamber (fig. 5B).

The trabecular pouch was posterior and to the left in six cases (figs. 1, 5A, 5B, 6A and 6B). In the other four cases, it was inferior and to the right (figs. 7A and 7B). The size of the trabecular pouch was highly variable, and was rather small in seven cases. A common denominator for all the cases was the angiographic appearance of the trabecular pouch walls, all of which were smooth and of a left ventricular pattern (figs. 5A, 5B, 7A and 7B). No papillary muscles were seen in the trabecular pouch.

Regarding the ventriculoarterial connections, the aorta and the pulmonary artery were present in all cases and originated from the main ventricular chamber, fulfilling the requirements of a double outlet right ventricle. The aortic valve was discontinuous from the atrioventricular valves in nine of the cases. The pulmonary valve was also discontinuous in nine cases. Therefore, in most cases, the arterial valves were supported by infundibular musculature separating them from the atrioventricular valves (figs. 7C and 7D).

The interrelation between the great arteries was as follows: In nine cases, the aorta and the pulmonary artery were at the same level in the craniocaudal plane; in the tenth case, the aorta was caudal to the pulmonary artery (fig. 5B). In the frontal plane, the aorta was anterior and to the left in five cases, anterior and to the right in four cases, and directly posterior in one case (table 1). The aortic arch was to the left in nine cases and to the right in the tenth case.

The most important associated anomalies in the series were: pulmonary stenosis — seven cases; atrial septal defect — six cases; left superior vena cava — one case; absence of right pulmonary artery — one case.

The angiographic pattern in this series was a densely opacified trabecular pouch and the great arteries in the presence of a poorly opacified main chamber during diastolic phase. This usually gives the appearance of two dense areas separated by a clear zone (figs. 6A, 6B, 7A and 7B). The main ventricular chamber thus cleared rapidly with the arrival of non-opacified blood from both atria. One case (patient 8) died after an attempt at surgical correction. The angiographic study (fig. 7) showed: 1) Solitus of the atra, 2) univentricular heart with two atrioventricular valves entering the morphological right ventricle, 3) trabecular pouch with left ventricular morphology located inferiorly and to the right, 4) double outlet right ventricle with aorta anterior and to the left, 5) pulmonary stenosis, and 6) atrial septal defect. A summary of the pathologic findings is as follows (fig. 8): 1) solitus of the atra, 2) large ventricular chamber with right ventricular morphology divided by the infundibular septum in two portions, 3) trabecular pouch with left ventricular morphology located inferior and to the right, 4) straddling left atrioventricular valve, 5) double outlet right ventricle, 6) well-developed ventriculo-infundibular fold under each arterial valve, and 7) aorta anterior and to the left.
Discussion

Different terminology has been used to define univentricular heart of the right ventricular type. Van Praagh et al. reported three cases of single ventricle with well-developed morphological right ventricle in absence of left ventricular sinus (type B). Muñoz-Castellanos called this malformation double inlet right ventricle and Quero-Jimenez, after an embryological study, identified it as an exaggerated displacement of the atrioventricular canal toward the bulbus cordis. Keeton et al. introduced the term univentricular heart of right ventricular type, pointing to the fact that the left ventricular chamber was directly comparable with the rudimentary chamber seen more frequently in “single ventricle with outlet chamber.” Previous reports of angiographic studies have failed to demonstrate this type of univentricular heart.

The angiographic identification of the anatomy in univentricular heart of right ventricular type has been facilitated in our last three cases by axial angiography developed by Bargeron et al. With this technique, the ventricular main chamber (right ventricular type) can be visualized in some aspects that are not easily seen in conventional frontal and lateral views, such as straddling valves, chordae tendineae, and trabecular pouch.

In three of our 10 cases, one atrioventricular valve was absent. These particular cases have been called by others tricuspid or mitral atresia, but we prefer to use the term of Anderson et al. and call them univentricular hearts, and specify which connection is absent.

The diagrams in figure 2 were made to clarify this complex cardiac malformation. We do not propose a new classification. Although there was no case with common atrioventricular valve in our series, we included it in the scheme because it is possible.

In the case in which an autopsy was performed, a very small and rudimentary papillary muscle was found in the trabecular pouch. This probably accounts for our failure to identify such structures in the cineangiography of this chamber. During cineangiography the trabeculated pouch remained opacified longer than the main ventricular chamber because of the relatively restricted septal defects that preclude free blood interchanges between the main chamber and trabecular pouch, and because, unlike the trabecular pouch, the nonopacified blood from the atria washes out the contrast media rapidly from the main chamber. The angiographic image at this stage is such that the opacified trabecular pouch and the opacified great arteries seemed to be separated from each other by the clear zone that represents the main ventricular chamber.

We believe that this type of univentricular heart can be diagnosed angiographically by its peculiar angiographic appearance. The main ventricular chamber that receives the two atrioventricular valves has the angiographic features of a normal right ventricular chamber and the presence of the trabecular pouch (rudimentary chamber) with left ventricular morphology makes certain the unequivocal diagnosis of univentricular heart of the right ventricular type.

Acknowledgment

We thank Dr. Robert H. Anderson from Brompton Hospital, London, England for his advice and critical appraisal of this manuscript.

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Angiographic study of univentricular heart of right ventricular type.
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Circulation. 1979;60:1325-1334
doi: 10.1161/01.CIR.60.6.1325
Circulation is published by the American Heart Association, 7272 Greenville Avenue, Dallas, TX 75231
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Print ISSN: 0009-7322. Online ISSN: 1524-4539

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