Congenital Malformations of the Right Atrium and the Coronary Sinus*

An Analysis Based on 103 Cases Reported in the Literature and Two Additional Cases

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Study objectives: Congenital malformations of the right atrium (RA) and the coronary sinus (CS) are rare, and only sporadic cases have been reported. Little is known about the clinical relevance of this disorder. We report on two patients, one with a giant RA diverticulum, the other with a diverticulum of the CS, and review 103 cases of such malformations that have been reported previously.

Design: A MEDLINE search was performed to collect all cases of congenital malformations of the RA and the CS reported in the literature between 1955 and 1998. Cases were classified into the following categories: (1) congenital enlargement of the RA; (2) single diverticulum of the RA; (3) multiple diverticula of the RA; and (4) diverticulum of the CS. Clinical presentation and outcome of the different types of malformations were analyzed.

Results: The patients most frequently presenting with symptoms were those with diverticula of the CS (n = 28) followed by those with single diverticula of the RA (n = 13), multiple diverticula (n = 4), and congenital enlargements of the RA (n = 60). The percentages of symptomatic patients were 93, 84, 75, and 53%, respectively. Symptoms were frequently caused by arrhythmias. Supraventricular tachycardia (SVT) was found in 42 of the patients (40%) and was most common in patients with diverticula of the CS (24 of 28 patients) and multiple atrial diverticula (3 of 4 patients). Sudden cardiac death was reported more frequently in patients with diverticula of the RA (5%) compared to those with congenital enlargement of the RA (5%) or single or multiple diverticula of the RA (6%). All seven patients with diverticula of the CS who were not treated with catheter or surgical ablation eventually died.

Conclusion: Congenital malformations of the RA and the CS frequently are associated with arrhythmias. SVT and sudden cardiac death have been reported in a significant percentage of patients with diverticula of the CS.

Key words: congenital heart disease; diverticulum of the coronary sinus; diverticulum of the midcardiac vein; right atrial aneurysm; right atrial diverticulum; right atrial enlargement; right atrium

Abbreviations: CS = coronary sinus; RA = right atrium/atrial; SVT = supraventricular tachycardia

The first case of congenital enlargement of the right atrium (RA) was described by Bailey in 1955. Since that time, RA malformations, including diffusely enlarged right atria, single or multiple saccular diverticula, or aneurysms arising from the RA or the coronary sinus (CS), have been reported repeatedly in the literature. However, since in most instances only single cases have been reported, little is known about the clinical relevance of those malformations and whether the distinct morphologies described differ with respect to clinical, diagnostic, and prognostic features. Since there have been reports of significant symptoms and even sudden death, this information is essential for correct diagnosis and appropriate patient management.2,8

We recently examined two patients: one had a giant RA diverticulum, and the other had a diverticulum of the CS. Based on these patients and an
extensive survey of the international literature, we sought to study the morphologic variety of such malformations, as well as possible differences in clinical presentation and patient outcome.

**Case Reports**

**Case 1**

A 40-year-old white man presented with atrial fibrillation and chest discomfort. A chest radiograph made during childhood had revealed a grossly enlarged heart, but no definite diagnosis had been made. The patient had never been seriously ill. An ECG revealed atrial fibrillation with a ventricular rate of 100 beats/min, right axis deviation, and nonspecific T-wave abnormalities. Chest radiography (Fig 1) showed a markedly enlarged cardiac silhouette. Laboratory findings were normal. Echocardiography demonstrated a huge (10 × 6 × 8 cm) cavity arising from the anterior free wall of the RA, superior to the tricuspid valve (Fig 2). This diverticulum extended cranial to the superior vena cava and the ascending aorta and caudal to the apex of the heart, where it surrounded the free right ventricular wall. Trabeculations, spontaneous echo contrast, and echoes that were characteristic of thrombotic material were present within the diverticulum. With the exception of mild tricuspid regurgitation with the regurgitant jet directed into the RA diverticulum, no other cardiac abnormalities or defects were noted. The morphologic aspects of the pathology were confirmed with MRI, which further demonstrated compression of the superior vena cava by the RA diverticulum (Fig 3). The patient was treated with warfarin, to prevent embolic events, and digoxin, for heart rate control. Atrial fibrillation persisted at a heart rate of 70 to 85 beats/min but was well tolerated. Surgical resection of the diverticulum was considered; however, the patient refused the operation. Thus, no further therapy was initiated. At follow-up 3 years after the initial presentation, the patient still reported palpitations but was otherwise free of symptoms.

**Case 2**

A 55-year-old woman with a history of recurrent orthodromic AV-nodal reentry tachycardia was admitted for electrophysiologic evaluation. The patient had a history of autoimmune thyroiditis; however, thyroid hormone levels were unremarkable. The number of episodes of supraventricular tachycardia (SVT) had increased over time to approximately one event per week. The results of a physical examination, ECG, chest radiograph, and laboratory tests were normal. The electrophysiologic study revealed an accessory pathway located in the posteroseptal region in close proximity to the CS. Retrograde angiography of the CS using a balloon occlusion catheter was performed and revealed a pulsatile, 3 × 3-cm diverticulum of the midcardiac vein (Fig 4). Successful high-frequency catheter ablation at the neck of the diverticulum was performed. Six months after the

**Figure 1.** Chest radiographs demonstrating a severely enlarged cardiac silhouette with a prominent right heart border.
procedure, the patient remains asymptomatic. In particular, no further episodes of SVT have occurred.

**Materials and Methods**

**Data Sources**

We performed a search of the international literature (MEDLINE) from 1955 through November 1998. Articles without individual case data and duplicate reports were excluded. In addition, cases with possible secondary causes of RA enlargement such as atrial septal defects, anomalous pulmonary venous return, Ebstein’s anomaly, and other lesions of the tricuspid or pulmonary valve, and pulmonary hypertension were excluded.

**Data Analysis**

The cases were classified into the following categories: (1) congenital enlargement of the RA; (2) congenital single diverticulum of the RA; (3) congenital multiple diverticula of the RA and (4) congenital diverticulum of the CS.

The following data were collected: (1) clinical information (age, gender, physical findings, symptoms, and associated congenital defects); (2) diagnostic modalities that were applied to establish the diagnosis; (3) diagnostic findings, in particular with respect to arrhythmias (ECG findings, the presence and type of arrhythmias, and the locations of accessory pathways); (4) morphologic characteristics (size and location of the malformation and presence of a thrombus); (5) the type and result of therapy and the reported outcome; and (6) the results of autopsy and histology.

**Statistical Analysis**

All values are presented as mean ± SD.

**Results**

**Data Sources**

The MEDLINE search revealed 141 patients reported in the literature. Thirty-eight patients had to be excluded. Thus, 103 patients that were reported on in 77 articles were analyzed. With the addition of the two cases from our institution, a total of 105 patients were studied.

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**Figure 2.** Transthoracic echocardiography showing the large RA diverticulum. *Left, A:* four-chamber view. *Right, B:* parasternal short axis view. Div = diverticulum; Ao = aorta; LA = left atrium; LV = left ventricle; RV = right ventricle.

**Figure 3.** MRI depicting the large diverticulum. *Top:* note how the diverticulum compresses the superior vena cava (VCS). *Bottom:* shows the communication between the diverticulum and the RA. Ao desc = descending aorta. Se Figure 2 for other abbreviations.
Classification

The most commonly reported type of malformation was a congenital enlargement of the RA (n = 60) followed by diverticulum of the CS (n = 28), single diverticulum of the RA (n = 13), and multiple diverticula of the RA (n = 4).

Patient Characteristics

The gender and age of the total population and the subgroups are shown in Table 1. Associated congenital malformations were present in 13 patients. Three patients had hypertrophic cardiomyopathy,9–11 five patients had ventricular septal defects12–14 (and one of these also had a hypoplastic left heart and a persistent left superior vena cava15), and one patient had a malformation of the bronchial system.16 A hamartoma of the liver was present in one patient,17 and one patient presented with a dilated urethropelvic system.5 Additional congenital abnormalities were most common in patients with a diverticulum of the CS (n = 7). Associated idiopathic enlargement of the left atrium was noted in seven patients (in six with enlarged RAs and one with multiple diverticula of the RA). There were five reports of familial occurrences of enlarged RAs.18

Clinical Presentation

Congenital enlargement of the RA and single diverticula of the RA were frequently accidental findings after recognition of an enlarged cardiac silhouette during routine chest radiography. In contrast, patients with diverticula of the CS and with multiple diverticula of the RA frequently presented with symptomatic tachyarrhythmia (82% and 75%, respectively).

Further evaluation revealed that only 31% of all reported patients were, indeed, found to be asymptomatic, with the highest percentage in the patient group with congenital enlargement of the RA (48%). Most frequently, patients reported arrhythmias and palpitations followed by shortness of breath and chest discomfort (Table 2).

ECG Findings

The ECG findings at presentation for the entire study population and the various subgroups are summarized in Table 3. Fifty-two patients (49%) were in sinus rhythm, and 29 patients (28%) were in atrial fibrillation or atrial flutter. Twelve patients (11%) presented with incessant SVT. All but two of these patients had either a diverticulum of the CS (seven patients) or multiple diverticula of the RA (three patients). A total of 42 patients had histories of, presented with, or developed SVT (paroxysmal or incessant). An accessory pathway was found in 25 of these patients. SVT and accessory pathways were much more common in patients with diverticula of the CS than in the other subgroups (Table 4). Of the 16 patients in whom the location of the pathway could be determined, 15 (93%) were posteroseptal and 1 was left sided.

Table 1—Patient Characteristics

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>All</th>
<th>C Enlarg RA</th>
<th>Single Div RA</th>
<th>Multiple Div RA</th>
<th>Div CS</th>
</tr>
</thead>
<tbody>
<tr>
<td>Patients, No.</td>
<td>105</td>
<td>60</td>
<td>13</td>
<td>4</td>
<td>28</td>
</tr>
<tr>
<td>Age, yr</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>28.4</td>
<td>28.9</td>
<td>30.6</td>
<td>19.0</td>
<td>27.4</td>
</tr>
<tr>
<td>Minimal</td>
<td>79</td>
<td>75</td>
<td>61</td>
<td>50</td>
<td>79</td>
</tr>
<tr>
<td>Gender, No.</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>62</td>
<td>36</td>
<td>6</td>
<td>3</td>
<td>17</td>
</tr>
<tr>
<td>Female</td>
<td>40</td>
<td>22</td>
<td>7</td>
<td>1</td>
<td>10</td>
</tr>
<tr>
<td>Unknown</td>
<td>2</td>
<td>2</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
</tbody>
</table>

*C = congenital; Div = diverticulum; Enlarg = enlargement; gest = gestational.
Diagnostic Techniques

**Congenitally Enlarged RA and Diverticulum of the RA:** Even though a chest radiograph often raised the suspicion of a cardiac malformation in these patients, the diagnosis usually was established with other techniques such as angiography (in the earlier years), echocardiography, CT, MRI, fetal ultrasound, and thoracotomy. Congenital Multiple Diverticula of the RA: The four patients with multiple diverticula of the RA had their conditions diagnosed in the following ways: by echocardiography in one patient, intraoperatively, as an accidental finding during cardiac surgery, in two patients, and by angiography in one patient. Cardiomegaly was noted on the chest radiographs of three patients.

**Congenital Diverticulum of the CS:** Congenital diverticula of the CS were detected by retrograde angiography of the CS during electrophysiologic studies in 20 of the 28 patients (71%). The remaining patients had their conditions diagnosed in the following ways: four patients at autopsy, three patients with transthoracic echocardiography, and one intraoperatively. Abnormal chest radiographs were seen only in four patients (14%).

**Diagnostic Pitfalls**

Three patients initially were suspected to have Ebstein’s anomaly. A tumor was suspected in one patient, pericardial effusion in two patients, and pericardial cysts in three patients.

**Morphologic Characteristics**

**Congenital Enlargement of the RA:** The enlarged RA was described as being “paper thin” and translucent in several patients. The largest right atria were those reported by De Marco and Bollero, with volumes of 900 cm³, and Saigusa et al, who described the RA to be as large as the “head of a child.” Progressive enlargement of the RA was reported in four patients. Tricuspid regurgitation secondary to congenital enlargement of the RA was found in three patients.

**Single Diverticulum of the RA:** Single diverticula of the RA usually were described as saccular structures originating from the free atrial wall (n = 10) or the RA appendage (n = 3). The size of the diverticulum varied considerably. The largest diverticulum was the one described by us (10 × 6 × 8 cm). The diverticula communicated with the RA via a broad neck in all patients. Köhne and Wellens described the wall of the diverticula as thin, while Sheldon et al reported that a thick layer of lipomatous tissue covered the aneurysmal sack.

**Multiple Diverticula of the RA:** Congenital multiple diverticula of the RA comprised three diverticula in two patients, and five diverticula in one patient. One patient had numerous saccular struc-

### Table 3—ECG Findings*

<table>
<thead>
<tr>
<th>Rhythm</th>
<th>All</th>
<th>C Enlarg RA</th>
<th>Single Div RA</th>
<th>C Multiple Div RA</th>
<th>Div CS</th>
</tr>
</thead>
<tbody>
<tr>
<td>SR</td>
<td>52 (49)</td>
<td>32 (53)</td>
<td>9 (69)</td>
<td>1 (25)</td>
<td>10 (35)</td>
</tr>
<tr>
<td>Atrial fibrillation</td>
<td>29 (25)</td>
<td>17 (28)</td>
<td>2 (15)</td>
<td>0 (0)</td>
<td>10 (37)</td>
</tr>
<tr>
<td>Intermittent atrial fibrillation</td>
<td>2 (2)</td>
<td>1 (2)</td>
<td>1 (8)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>AV junction rhythm</td>
<td>4 (4)</td>
<td>4 (7)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>AV block</td>
<td>2 (2)</td>
<td>2 (3)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Incessant SVT</td>
<td>12 (11)</td>
<td>2 (3)</td>
<td>0 (0)</td>
<td>3 (75)</td>
<td>7 (26)</td>
</tr>
<tr>
<td>Asystole</td>
<td>1 (1)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>1 (4)</td>
</tr>
<tr>
<td>Unknown</td>
<td>3 (3)</td>
<td>2 (3)</td>
<td>1 (8)</td>
<td>0 (0)</td>
<td>0 (0)</td>
</tr>
</tbody>
</table>

*Values given as No. of patients (%). SR = sinus rhythm. See Table 1 for other abbreviations.
tures, which covered the entire RA wall.\textsuperscript{23} These diverticula were considerably smaller than single diverticula of the RA, the largest being 3.2 cm in diameter.\textsuperscript{24} Miyamura et al.\textsuperscript{24} reported that the diverticula showed deposits of lipomatous tissue and fatty infiltration.

\textit{Diverticula of the CS:} Most of the diverticula arose directly from the CS (n = 24), while four originated from the midcardiac vein.\textsuperscript{3,12,39} The largest diverticulum was 5 \times 3 cm.\textsuperscript{40} The wall of the diverticula often was composed of muscular bundles, and several diverticula, including ours, were pulsatile.\textsuperscript{12,41}

\textit{Thrombus:} A thrombus within the diverticulum or enlarged RA, diagnosed by imaging procedures, intraoperatively, or at autopsy, was present in only 4 of 73 patients.\textsuperscript{27,42} No patient with a diverticulum of the CS or with multiple diverticula of the RA had a thrombus. The largest thrombus described was the size of a tennis ball.\textsuperscript{42}

\textit{Treatment:} The different treatment modalities that were applied for the various subgroups are shown in Table 5. Surgical resections of diverticula or reductions of RA size were performed in 40 patients (38\%) and were performed most often in patients with multiple diverticula of the RA (100\%), followed by those patients with single diverticula of the RA (69\%), diverticula of the CS (36\%), and congenital enlargement of the RA (28\%). Almost half of these surgical procedures (19 patients) were performed to abolish the arrhythmogenic substrate (surgical ablation). In these patients, surgery was successful in 18 patients and unsuccessful (ie., there was a recurrence of arrhythmia) in 1.\textsuperscript{30} One patient with multiple diverticula successfully underwent surgery but suffered a cerebral embolism. The remaining procedures were performed to establish the diagnosis (exploration, 3 patients), because a pericardial cyst or mediastinal tumor was suspected (3 patients), or because it was believed that the patients would benefit in terms of outcome (16 patients). The operative technique usually involved resection of the diverticulum or partial resection of the aneurysmatic wall of the RA. These procedures were possible, with the exception of one patient with multiple diverticula,\textsuperscript{23} in whom only partial resection was possible. No operative deaths were reported.

Catheter ablation was performed successfully in all 12 patients with preexcitation syndrome (all associated with a diverticulum of the CS). Twenty patients received antiarrhythmic medication and 3 received warfarin, while 23 patients received no therapy.

\textit{Outcome and Follow-up Data:} Patient follow-up was reported in 43 patients (41\%), with a mean follow-up duration of 3.1 \pm 4.2 years (range, 6 wks to 20 years). Eleven deaths were reported (Table 6). Of those deaths, 10 were cardiac related, and 8 of these (72\%) occurred suddenly. Sudden cardiac death was reported most frequently in patients with diverticula of the CS (5 of 28 patients). Of the seven patients with diverticula of the CS that were not treated with catheter or surgical ablation, five died suddenly, one died as a consequence of hypoplastic left heart disease,\textsuperscript{15} and one died following a myocardial infarction.\textsuperscript{8}

### Table 4—Tachycardia Data\textsuperscript{*}

<table>
<thead>
<tr>
<th>Tachycardia</th>
<th>All</th>
<th>C Enlarg RA</th>
<th>Single Div RA</th>
<th>C Multiple Div RA</th>
<th>C Div CS</th>
</tr>
</thead>
<tbody>
<tr>
<td>SVT</td>
<td>42 (40)</td>
<td>11 (19)</td>
<td>4 (31)</td>
<td>3 (75)</td>
<td>24 (86)</td>
</tr>
<tr>
<td>Preexcitation</td>
<td>25 (24)</td>
<td>1 (2)</td>
<td>2 (15)</td>
<td>0 (0)</td>
<td>22 (79)</td>
</tr>
</tbody>
</table>

\textsuperscript{*}Values given as No. of patients (%). See Table 1 for abbreviations.

### Table 5—Treatment Data\textsuperscript{*}

<table>
<thead>
<tr>
<th>Treatment</th>
<th>All</th>
<th>C Enlarg RA</th>
<th>Single Div RA</th>
<th>C Multiple Div RA</th>
<th>C Div CS</th>
</tr>
</thead>
<tbody>
<tr>
<td>Operation</td>
<td>40 (38)</td>
<td>17 (28)</td>
<td>9 (69)</td>
<td>4 (100)</td>
<td>10 (36)</td>
</tr>
<tr>
<td>Catheter ablation</td>
<td>12 (11)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>12 (43)</td>
</tr>
<tr>
<td>PM</td>
<td>4 (4)</td>
<td>3 (5)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td>1 (4)</td>
</tr>
<tr>
<td>Medical</td>
<td>20 (19)</td>
<td>15 (25)</td>
<td>1 (8)</td>
<td>1 (25)</td>
<td>3 (11)</td>
</tr>
<tr>
<td>No therapy</td>
<td>23 (22)</td>
<td>19 (32)</td>
<td>3 (23)</td>
<td>0 (0)</td>
<td>1 (4)</td>
</tr>
<tr>
<td>Unknown</td>
<td>10 (10)</td>
<td>8 (13)</td>
<td>1 (8)</td>
<td>0 (0)</td>
<td>1 (4)</td>
</tr>
</tbody>
</table>

\textsuperscript{*}PM = pacemaker. See Table 1 for other abbreviations.
Discussion

There are four distinct types of malformations of the CS and the RA: congenital enlargement of the RA; single RA diverticulum; multiple diverticula of the RA; and diverticula of the CS. It is difficult to estimate the true incidence of the various subgroups since smaller diverticula, especially in asymptomatic patients, easily could be missed. Particularly for patients with diverticula of the CS in whom diagnosis generally requires angiography of the CS, the diagnostic procedure will be performed only in symptomatic patients undergoing electrophysiologic studies. Several authors report an incidence of 4 to 12% in patients with posteroseptal pathways, demonstrating that in this patient population, diverticula of the CS are more common than previously suspected.

There appears to be no gender or race preferences, and although there is a report of a familial occurrence, which could suggest a genetic predisposition, this finding has not been confirmed. Associated congenital anomalies are seen most frequently in patients with diverticula of the CS.

The clinical presentation and diagnostic approach greatly depend on the type of malformation that is present. Patients with congenital enlargement of the RA and those with large diverticula of the RA frequently come to medical attention because of a gross enlargement of the cardiac silhouette on chest radiographs or because of atrial arrhythmias. In contrast, patients with diverticula of the CS usually present with SVT associated with accessory pathways that transverse the diverticulum to form an atrioventricular connection.

Patients with multiple diverticula of the RA frequently present with incessant atrial tachycardia that most likely are caused by an ectopic pacemaker focus located in the vicinity of the diverticula. These arrhythmias usually occur early in life during infancy or childhood.

Large diverticula can cause chest discomfort and symptoms related to compression of intrapericardial cardiac structures such as jugular engorgement, edema of the ankles, and hepatomegaly. Congestive heart failure rarely occurs and is usually related to impaired systolic left ventricular function caused by incessant tachycardia. Some patients with enlarged RAs develop secondary tricuspid regurgitation due to dilatation of the tricuspid annulus. However, many patients, in particular, those with single diverticula and enlarged RAs, remain asymptomatic until late in life.

In the modern era, the diagnosis usually is established with echocardiography (transthoracic, transesophageal, and fetal), angiography of the CS, MRI, or CT. These modalities also help to exclude a pericardial cyst or a mediastinal tumor, which can mimic congenital malformations of the RA. Because diverticula of the CS are closely linked to patients with posterior accessory pathways and because these anomalies have important implications for ablative procedures, routine angiography of the CS in such patients has been suggested.

The wall of the enlarged RA and of RA diverticula often demonstrates lipomatous degeneration and reduction of muscular elements. In contrast, diverticula of the CS are composed of muscular bundles running within the wall and are often contractile.

It is difficult to assess the prognosis of patients with such malformations since follow-up is not available in many instances. Eleven deaths have been reported in the literature. However, at least five of these deaths can be attributed either to other causes or to associated defects.

Nevertheless, there is evidence in the literature that untreated symptomatic patients with diverticula of the CS are at high risk for cardiac death. This risk may be due to the frequently encountered short,
antegrade refractory periods of such accessory pathways, which predispose patients to rapid ventricular responses in atrial fibrillation.14,41 However, it remains unclear whether the high mortality rate in these patients is related to a selection bias.

The optimal therapeutic approach is still controversial. Catheter ablation of accessory pathways in patients with diverticula of the CS appears to be the treatment of choice in symptomatic patients.40,53–57

The low operative mortality rate and high success rate justify a surgical approach in patients with multiple and single diverticula of the atria and SVT. However, in the absence of symptomatic arrhythmias, most authors suggest that surgery should be performed only in patients with symptomatic right ventricular compression, while asymptomatic patients should be treated conservatively. Because patients are at increased risk of developing a thrombus in the RA (particularly patients with diverticula of the RA) and because of the high likelihood of developing atrial fibrillation, such patients may require anticoagulation.

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