

“Crochetage” (Notch) on R Wave in Inferior Limb Leads: A New Independent Electrocardiographic Sign of Atrial Septal Defect

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Objectives. This study sought to determine the clinical significance of a “crochetage” pattern—a notch near the apex of the R wave in electrocardiographic (ECG) inferior limb leads—in secundum atrial septal defect.

Background. Atrial septal defect is often overdiagnosed on the basis of classical clinical features. Thus, more specific signs on the ECG for screening are needed.

Methods. We searched for a crochetage pattern in 1,560 older children and adults: 532 with secundum atrial septal defect, 266 with ventricular septal defect, 146 with pulmonary stenosis, 110 with mitral stenosis, 47 with cor pulmonale and 459 normal subjects.

Results. This pattern was observed respectively in 73.1%, 35.7%, 23.3%, 6.4%, 10.6% and 7.4% of these groups ($p < 0.001$). In atrial septal defect, its incidence increased with larger anatomic defect ($p < 0.0001$) or greater left-to-right shunt ($p < 0.0001$), even in

the presence of pulmonary hypertension. By multiple regression analysis, only shunt size ($p < 0.0006$) and defect location ($p < 0.0001$) were the determinants of its presence. In all groups, the specificity of this sign for the diagnosis was remarkably high when present in all three inferior limb leads ($\geq 92\%$), even when comparison was limited to patients with an incomplete right bundle branch block ($\geq 95.2\%$). Early disappearance of this pattern was observed in 35.1% of the operated-on patients although the right bundle branch block pattern persisted.

Conclusions. A crochetage pattern of the R wave in inferior limb leads is frequent in patients with atrial septal defect, correlates with shunt severity and is independent of the right bundle branch block pattern. Sensitivity and specificity of this sign are remarkably high when it is associated with an incomplete right bundle branch block or present in all inferior limb leads.

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The main consequences of atrial septal defect are the appearance of pulmonary hypertension and congestive heart failure, but there is also a potential for transitory right-to-left shunting with its potential for cerebral paradoxical embolism and arterial desaturation such as encountered in scuba divers (1-3). Some may then suggest that all atrial septal defects, even those clinically silent, would have to be closed (1,4-7). In this setting, an incomplete right bundle branch block pattern on the electrocardiogram (ECG) may often orient toward an echocardiographic examination but is lacking in specificity (1,8-11). On the contrary, a notched aspect of the R wave of the QRS complex in inferior ECG limb leads was frequently observed in our practice in ostium secundum or sinus venosus atrial septal defects and seemed independent of the right bundle branch block. This pattern was called “crochetage,” a French equivalent of notch, in 1959 in a report of a group of 11 patients with secundum atrial septal defect (12), but the clinical relevance of this pattern remained unknown. We studied its clinical signifi-

cance in a large cohort of patients with atrial septal defect and tested its diagnostic value in a normal group and in groups with other causes of incomplete right bundle branch block and right ventricular hypertrophy as in ventricular septal defect, pulmonary stenosis, mitral stenosis or cor pulmonale.

Methods

Patients. *Atrial septal defect study group.* A total of 568 older children and adults with atrial septal defect underwent catheterization in our institution between January 1960 and December 1984. We excluded patients with ostium primum defect or with other associated hemodynamically severe congenital heart disorder or patients with anomalous venous drainage without atrial septal defect; patients with partial anomalous pulmonary venous drainage associated with an atrial septal defect were included in the study. Thus, 532 patients (374 female) were retrospectively included in this study. Mean age was 24.1 ± 15.0 years (range 4 to 80). All patients underwent clinical and ECG examination; 50 patients presented with clinical signs of congestive right heart failure.

The diagnosis was confirmed during catheterization by passage of the catheter across the defect at a midseptal level or by an increase of 10% in oxygen saturation of right atrial blood compared with blood from the superior and inferior venae cavae. In all cases, mean pulmonary artery pressure and the

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magnitude of left-to-right shunting across the defect (pulmonary/systemic blood flow ratio: Q_p/Q_s) were measured using the direct Fick method. Mean pulmonary artery pressure was 21.3 ± 13.9 mm Hg (range 6 to 130); pulmonary hypertension, defined as mean pulmonary artery pressure >30 mm Hg, was present in 83 patients (15.6%). Mean Q_p/Q_s was equal to 3.47 ± 2.12 (range 0.4 to 8.1), and the shunt was considered significant when the Q_p/Q_s ratio was >1.5 . Contraindications to operation—severe pulmonary hypertension, severe associated diseases or old age—were found by cardiologist referees in 48 cases; in this group, mean age was 42.3 ± 19.0 years, (range 6 to 80) and mean pulmonary artery pressure was 54.6 ± 27.3 mm Hg (range 30 to 101).

Surgical repair of the defect was performed in 484 patients (91%) (336 female) with a mean age of 22.6 ± 13.5 years (range 4 to 72); 35 patients (7.1%) with moderate pulmonary hypertension and high level shunting across the defect underwent operation. This made it possible to confirm the type of ostium secundum defect (13) and the associated lesions: central type (58.9%); inferior caval type or low septal defect (19.2%); high septal or sinus venosus defect (15.3%) associated with a partial anomalous venous drainage in 50 cases; large defect from the superior to the inferior vena cavae (6.6%), associated with a partial anomalous venous drainage in 2 patients.

Congenital heart disease control group. This group included 412 consecutive patients admitted during the same period of time and without complete right bundle branch block pattern. There were 266 patients (164 male) with isolated ventricular septal defect (mean age 14.3 ± 18 years [range 6 to 38]) and 146 patients (91 male) with isolated pulmonary stenosis (mean age 19.1 ± 15 years [range 8 to 42]). The diagnosis was based on Doppler and echocardiographic examinations or heart catheterization and confirmed by surgical repair in 305 patients.

Acquired heart disease control group. This group included 157 consecutive patients admitted during the same period of time and without a complete right bundle branch block pattern. There were 110 patients (75 female) with isolated or predominant severe mitral stenosis (mean age 58.3 ± 22.1 years [range 18 to 62 years]); mean pulmonary artery pressure as assessed by catheterization was 32.3 ± 25.7 mm Hg (range 16 to 50), and operation was performed in 104 cases. There were 47 patients (32 male) with cor pulmonale from chronic hypoxia, pulmonary embolism or primary pulmonary hypertension (mean age 68.1 ± 17.9 years [range 49 to 82]); mean pulmonary artery pressure as assessed by Doppler examination or right heart catheterization was 47.3 ± 20.7 mm Hg (range 39 to 70).

Normal control group. This group included 459 consecutive patients (mean age 24.3 ± 14 years, range 3.5 to 83; 227 female) admitted in our hospital for orthopedic or plastic surgery between 1960 and 1980 and who were considered to have a normal heart. These patients had no history of heart disease, no cardiovascular treatment, normal cardiovascular examination and chest radiograph; the ECG findings were

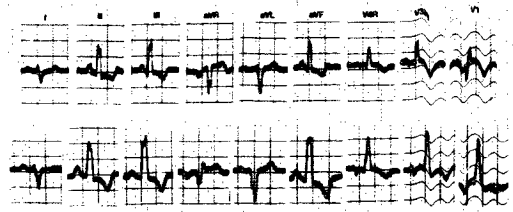


Figure 1. Examples of electrocardiographic (ECG) tracings with "crochetage" patterns in inferior limb leads II, III and aVF in patients with atrial septal defect. **Top,** ECG from an 11-year old girl with a central defect (mean pulmonary artery pressure was 17 mm Hg, and Q_p/Q_s was 3.8). Note the notch on the QRS near the zenith of the R wave or on its ascending branch and the incomplete right bundle branch block pattern in V_1 lead. **Bottom,** ECG from a 44-year old woman with a large defect between the venae cavae (mean pulmonary artery pressure was 40 mm Hg, and Q_p/Q_s was 1.4). Note the notch on the R wave in the inferior limb leads and the complete right bundle branch block pattern on the V_1 lead.

normal or showed a pattern of incomplete right bundle branch block without sign of right ventricular hypertrophy.

Electrocardiographic patterns. All patients had a standard 12-lead ECG with a sensitivity of 10 mm/mV and a 25 mm/s paper speed. Analysis of the tracings were performed in 100 patients in each of the four groups by two independent observers (J.H., A.H.) who were unaware of the clinical, hemodynamic and surgical data. Right bundle branch block on the ECG was defined as complete when QRS duration was >120 ms in adults with a r' or R' aspect in right precordial lead V_1 or V_2 and with a wide S wave (longer than R duration or than 40 ms) in leads V_6 and I; it was defined as incomplete when QRS duration was between 80 and 120 ms with a R' or r' in lead V_1 or V_2 and R' greater than R in V_1 or V_2 leads, or when QRS duration was <120 ms with an R peak time >70 ms in lead V_1 or V_2 (14). The QRS aspect on the preoperative ECG helped to define patients along with the absence or the presence of a "crochetage" aspect of the R wave in one or more of limb leads II, III or aVF (Fig. 1). This pattern—a notch on the R wave—was defined as a rapid upward-down motion of the R wave tracing on its ascending branch or near its zenith with an M-shaped or a bifid pattern in the most typical form and always involving the initial 80 ms of the QRS complex. Postoperative ECGs were obtained in all patients after atrial septal defect repair within 15 days after operation.

Statistical analysis. The data were analyzed by the SAS (SAS Institute, Inc., version 6.03). All results are expressed as mean value \pm SD. For ECG interpretation, concordance between the two observers was calculated. The Student's *t* test was used to assess differences in continuous variables between two groups, and one-way analysis of variance (ANOVA) was used to define a global statistical significance for differences between more than two groups. When a global statistical significance was obtained, a post hoc test (Tukey's standardized range test) was used to test for significant difference between two groups. A chi-square test (or a Fisher exact test) was used

to assess differences in categorical variables. Pearson correlation coefficient was used to study the relation between two quantitative variables. Multivariate analysis was used to search for factors related to the presence of a "crochetage" pattern and was performed using logistic regression analysis; when more than two classes were present for a categorical variable, a dummy variable was generated; parameters were estimated using the maximum likelihood method. Results were considered significant if $p < 0.05$.

Results

Mean age was not significantly different between the atrial septal defect and the normal groups but was lower in the congenital heart disease control group and higher in the acquired heart disease control group ($p < 0.01$).

Electrocardiographic data. Concordance between the two observers for ECG interpretation—presence of a right bundle branch block pattern and number of "crochetage"—was 99%.

Standard ECG data. All atrial septal defect group patients were in sinus rhythm, except for 10 in atrial fibrillation; PR interval duration was within the normal range (120 to 200 ms) in 94.9% (505 patients), >200 ms in 14 patients and >120 ms in 3 patients; 522 patients (98.1%) showed a right bundle branch block, incomplete in the majority of the patients (491 patients, 94.1%) and complete in the remaining patients (31 patients, 5.9%). In the normal group, an incomplete right bundle branch block was observed in 50 subjects (10.9%) without any other ECG abnormality, and its sensitivity and specificity for the diagnosis of atrial septal defect reached, respectively, 98.1% and 89.1%. Among patients with ventricular septal defect or pulmonary stenosis, an incomplete right bundle branch block was observed in 112 (27.2%), and its specificity for the diagnosis of atrial septal defect reached 71.1%. Among patients with mitral stenosis or cor pulmonale, an incomplete right bundle branch block was observed in 42 (26.8%), and its specificity for the diagnosis of atrial septal defect reached 73.2%.

Electrocardiographic "crochetage" pattern. Among atrial septal defect patients, a "crochetage" was observed in 73.1% of patients in one lead at least, 58.1% in two to three leads and 27.8% in three leads (Table 1); it was isolated (without right bundle-branch block) in three patients (Fig. 2). In the normal group, a crochetage was present in only 7.4% of the patients in at least one lead, but a crochetage in all inferior limb leads was never observed. According to these results in the normal control group, its sensitivity and specificity for the diagnosis reached, respectively, 73.1% and 92.6% if present in one inferior limb lead only, 58.1% and 97.2% if present in two to three leads and 27.8% and 100% if present in three inferior limb leads. Among patients with ventricular septal defect or pulmonary stenosis, a crochetage was observed, respectively, in 35.7% and 23.3% in one lead at least, in 17.3% and 8.9% in two to three leads and in 1.9% and 0.7% in three leads; thus, the sensitivity and specificity of the presence of a crochetage

Table 1. Frequency of "Crochetage" Pattern in Inferior Limb Leads in Study Patients

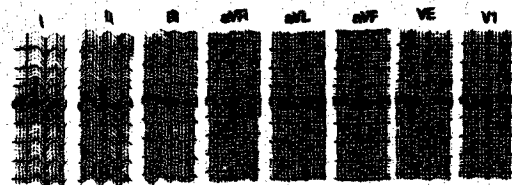
	One Lead	Two Leads	Three Leads
ASD	15.0% (80)	30.3% (161)	27.8% (148)
VSD	18.4% (49)	15.4% (41)	1.9% (5)
PS	14.4% (21)	8.2% (12)	.7% (1)
MS	2.7% (3)	1.8% (2)	1.8% (2)
CP	6.4% (3)	2.1% (1)	2.1% (1)
NLS	4.6% (21)	2.8% (13)	0% (0)

Values presented are percent (number) of patients. ASD = atrial septal defect; CP = cor pulmonale; MS = mitral stenosis; NLS = normal subjects; PS = pulmonary stenosis; VSD = ventricular septal defect.

pattern in three leads reached, respectively, 27.8% and 98.5%. Among patients with mitral stenosis or cor pulmonale, a crochetage was observed, respectively, in 6.4% and 10.6% in one lead at least, in 3.6% and 4.3% in two to three leads and in 1.8% and 2.1% in three leads; thus, in this control group of patients with acquired heart disease, potentially a cause of right ventricular hypertrophy, the sensitivity and specificity of the presence of a crochetage in three leads reached, respectively, 27.8% and 92.4%.

In the normal control group, a crochetage was associated with an incomplete right bundle branch block in only 5 patients (1.0%) and were not statistically linked ($p = 0.68$); a crochetage without right bundle branch block was observed in 29 patients. Similarly, in the congenital and acquired heart disease control groups, they were scarcely associated, respectively, in 66 patients (16%) and in 5 patients (3.2%); a crochetage without incomplete right bundle branch block was observed in these two groups, respectively, in 63 and 7 patients. When only patients with an incomplete right bundle branch block pattern were considered, the sensitivity for the diagnosis of atrial septal defect of a crochetage pattern reached, respectively, 70.3%, 57.4% and 26.9% when present in one lead at least, in two or three leads and in three inferior leads. Similarly, when only patients with a right bundle branch block pattern were considered in the normal, the congenital and the acquired control groups, its specificity for the diagnosis reached, respectively, 90%, 41.1%, 85.7% when present in one lead only, 94%, 66.7% and 91.5% when present in two to three leads, and 100%, 96.4% and 95.2% when present in all three inferior leads.

Figure 2. Example of electrocardiographic tracings with "crochetage" patterns in inferior limb lead III but without right bundle branch block pattern in a 79-year old man with secundum atrial septal defect (mean pulmonary artery pressure was 15 mm Hg, and Q_p/Q_s was 1.8).



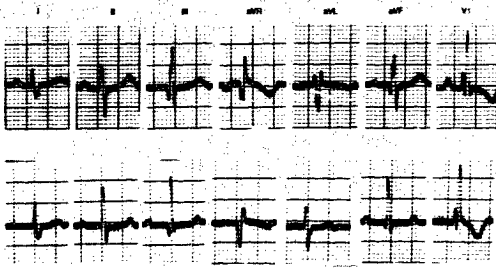


Figure 3. Electrocardiographic tracings from a 16-year old girl with a low septal defect and partial anomalous venous return. Preoperative mean pulmonary artery pressure was 15 mm Hg, and the Q_p/Q_s ratio was 2.1. Note the "crochetage" on the R wave tracing in inferior limb leads II, III, aVF and the incomplete bundle branch block pattern in the V₁ lead on the ECG before operation (top). Three days after atrial septal defect surgical repair, note the disappearance of the crochetage pattern, whereas the incomplete right bundle-branch block pattern persisted (bottom).

Effect of atrial septal defect repair on the ECG. Within 10 to 15 days (mean 12.6 days) after defect repair, the disappearance of the crochetage was observed in 191 patients (53.9%) among the 354 with this preoperative pattern in one lead at least; this was noted in 30 (45.5%) of the 66 patients with this pattern in one lead only and in 161 (55.9%) of the 288 patients with this pattern in two to three leads. Meanwhile, among these patients, the incomplete right bundle branch block persisted in 120 patients (63.2%); this was noted in 19 (65.5%) of the 29 patients with this pattern in one lead only preoperatively and in 101 (62.7%) of the 161 with this pattern in two to three leads preoperatively. Moreover, among the 288 patients with a preoperative crochetage pattern in two to three leads, 101 (35.1%) no longer had crochetage after operation but demonstrated persistence of an aspect of incomplete right bundle branch block that may disappear later (Fig. 3). Conversely, when the crochetage pattern persisted postoperatively (163 patients), the right bundle branch block pattern was no longer seen in 38.0% of patients (62 patients). Rarely, these two patterns may disappear simultaneously; this was observed in 70 patients (19.8%) of the 353 who presented preoperatively with these two patterns.

Hemodynamic data. Among patients with atrial septal defects, Q_p/Q_s was 3.9 ± 2.1 , 3.1 ± 1.4 and 1.9 ± 1.8 , respectively, in patients with operation without and with pulmonary hypertension and in patients without operation ($p < 0.01$). Patients with operation presented with a higher mean pulmonary artery pressure (54.6 vs. 18.0 mm Hg) ($p < 0.0001$) and older mean age (42.3 vs. 22.6 years) ($p < 0.0001$) than those without operation. Mean pulmonary artery pressure increased with age ($r = 0.41$, $p < 0.0001$), whereas Q_p/Q_s decreased with age ($r = -0.11$, $p < 0.01$) and with mean pulmonary artery pressure ($r = -0.18$, $p < 0.0001$).

Shunt severity and crochetage pattern. By univariate analysis, Q_p/Q_s was significantly different between groups with no

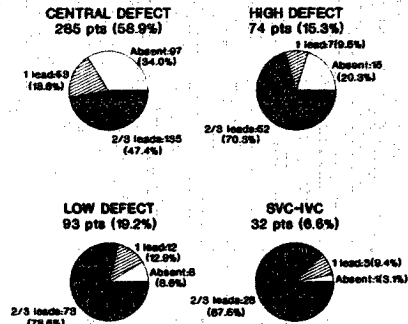


Figure 4. Frequencies of a "crochetage" pattern in inferior limb leads in 484 patients (pts) operated on and with different types of secundum atrial septal defect. The total number and percent of patients in each group are given according to the total number of patients with atrial septal defect. For each type of defect, the number of patients and percent within the group are given according to the presence of a crochetage pattern. Absent = no crochetage; 1 lead = presence of a crochetage in one lead only; 2/3 leads = presence of a crochetage in two or three leads. SVC-IVC = large defect from superior to inferior venae cavae.

crochetage (2.9 ± 1.9), crochetage in one lead only (3.6 ± 2.6) and crochetage in two to three inferior limb leads (3.8 ± 1.9) ($p < 0.0001$). Mean Q_p/Q_s was significantly higher in patients from the latter two groups ($p < 0.001$). Moreover, the presence of a crochetage in at least one lead differed significantly according to anatomic type of the defect: 65.9% for a central-type defect, 79.7% for a high septal defect, 91.4% for a low septal defect and 96.9% for a large defect between the venae cavae ($p < 0.001$) (Fig. 4). Similarly, Q_p/Q_s also differed significantly with the location of the defect ($p = 0.008$) and was more important for larger (between venae cavae) than for smaller (central-type) defects (4.5 ± 3.2 vs. 3.4 ± 1.8 , $p = 0.04$). Mean pulmonary pressure did not differ between groups with or without crochetage (22.8 ± 15.1 mm Hg in the absence of crochetage, 20.5 ± 13.9 mm Hg if present in one lead and 20.7 ± 13.3 mm Hg if present in two to three leads, $p > 0.05$) or with the location of the defect.

By multivariate analysis in the atrial septal defect group, only Q_p/Q_s ($p = 0.0006$) and anatomic type of the defect ($p < 0.0001$), but not age ($p = 0.57$), gender ratio ($p = 0.11$) or mean pulmonary artery pressure ($p = 0.08$), were independent determinants of the presence of a crochetage pattern in one inferior limb lead at least.

Despite pulmonary hypertension (83 patients), a crochetage in at least two inferior limb leads was observed in only 29% of the patients with a $Q_p/Q_s < 1.5$ versus 67.3% of patients with $Q_p/Q_s > 1.5$ ($p < 0.02$) (Fig. 5). In patients without operation with severe pulmonary hypertension (48 patients), a crochetage pattern was present in only 38.5% of those with $Q_p/Q_s < 1.5$ versus 72.7% of those with $Q_p/Q_s > 1.5$ ($p = 0.021$). For these patients, Q_p/Q_s was 1.3 ± 0.5 in patients without versus 2.3 ± 1.9 in those with crochetage ($p = 0.023$).

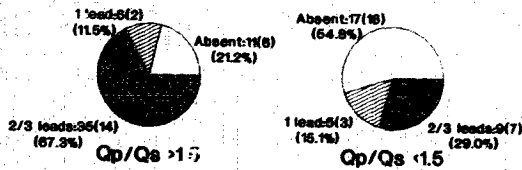


Figure 5. Number of patients with a "crochetage" pattern in inferior limb leads according to the magnitude of the left-to-right shunt in 83 patients with atrial septal defect and pulmonary hypertension (mean pulmonary artery pressure >30 mm Hg). Among them, the number of patients who did not undergo operation are shown in parentheses. Q_p/Q_s = pulmonary/systemic blood flow ratio; other abbreviations in Figure 4.

Discussion

Atrial septal defect, even if it is clinically silent, may be associated with nonthrombotic cerebral vascular accident in young patients (1,4,6). Conversely, diagnosis and early repair of atrial septal defect is associated with excellent long-term survival (1,3,15). However, most young patients with atrial septal defect have no symptoms, and, because of the nonspecific nature of the presenting symptoms and the relatively subtle physical signs, it is not unusual for atrial septal defect to be diagnosed first during early to middle adulthood or even older age (1,16). This emphasizes the need for a high electrocardiographic index of suspicion for the defect in all patients with compatible presenting signs and symptoms; the diagnosis may be then confirmed by echocardiographic techniques (1,17). The electrocardiogram in atrial septal defect can reveal supraventricular arrhythmias, QRS axis deviation, prolonged P-R interval, and right bundle branch block considered as one hallmark of the disease and present in our study in more than 90% of the cases (1,8-11). However, according to the literature, similar patterns may be seen in normal subjects in up to 2.9% to 5% of the cases (18,19); in the current study, this frequency is even higher reaching 10.9%.

An early notch on the R wave of the QRS in inferior limb leads had been reported 35 years ago in a small group of patients with an ostium secundum defect (11). This triphasic aspect, called "crochetage," was observed in our practice in at least one inferior limb lead in a majority of patients with atrial septal defect but in only 7.4% of normal subjects and in fewer than one-third of the patients with other congenital heart disease and one tenth of the patients with acquired heart disease. This frequent finding is a sensitive sign for the diagnosis of secundum atrial septal defect but no more so than incomplete right bundle branch block. However, when present in all three inferior limb leads, its specificity is considerably high (92% to 100%), even in populations with other causes of right ventricular hypertrophy such as in congenital (ventricular septal defect, pulmonary stenosis) or acquired (mitral stenosis, core pulmonale) heart diseases; this has to be compared with the specificity of the right bundle branch block pattern in such populations (71.1% and 73.2%, respectively, in patients with congenital and acquired heart diseases). Moreover, when

statistical comparison is limited to patients with an incomplete right bundle branch block pattern, its specificity, when present in all three inferior limb leads, remains considerably high; in all populations, greater than 95.2%.

In atrial septal defect, this pattern is clearly independent of the incomplete right bundle branch block: 1) it may be present in the absence of the right bundle branch block, as observed in several patients with atrial septal defect; 2) in the immediate postoperative period, it frequently disappears while the right bundle branch block, which has been attributed to chronic right ventricular volume overload, usually persists longer; 3) it always involves the initial 80 ms of the QRS, while the conduction disturbance on the right bundle branch involves the last part of the QRS wave. This "crochetage" is readily apparent in inferior limb leads and may be associated with a "crochetage" pattern in other limb leads; however, after surgery, crochetage patterns in all inferior standard leads disappear at the same time, although crochetage may persist in other standard leads.

The association between this triphasic pattern and left-to-right shunting is confirmed by the strong correlation between the presence of a crochetage pattern and shunt severity or anatomic type and size of the defect. Even in the case of pulmonary hypertension, presence of a crochetage pattern remains an index of significant left-to-right shunt and could lead to consideration of surgical repair of the defect.

Limitations of the study. Selection bias may have occurred in the atrial septal defect group, as only patients who underwent catheterization were included in the study; thus, more benign atrial septal defects with the smallest shunts may not have been studied; however, these patients usually have no indication for further explorations, as surgical repair is usually not indicated. Patients in the normal and atrial septal defect groups were not matched for age, but mean age was not different between these groups, and adjustment for age was performed using multivariate analysis. Finally, the exact cause of this pattern remains unknown even if it is related to the importance of the shunt; it might not be related to right ventricular hypertrophy and dilation, which often resolve 6 months to 1 year after operation, whereas the abnormality in right ventricular conduction seen on the ECG can resolve later (1,15).

Conclusions. A "crochetage" pattern on the R wave in inferior ECG limb leads is frequently seen in atrial septal defect patients and is independent of the frequent incomplete right bundle branch block pattern. The association of these two patterns considerably increases the specificity of the electrocardiogram for the diagnosis of an atrial septal defect. Though its pathophysiologic mechanisms remain unknown, the presence of this crochetage correlates with shunt severity, even in the presence of pulmonary hypertension.

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