

The Parasternal Short-Axis View Improves Diagnostic Accuracy for Inferior Sinus Venosus Type of Atrial Septal Defects by Transthoracic Echocardiography



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Background: Sinus venosus defects (SVD) of the inferior vena cava (IVC) type, or inferior SVDs, are an uncommon form of atrial communication located outside the confines of the fossa ovalis and involve override of the IVC. Despite numerous studies describing the anatomical and echocardiographic features of the inferior SVD, distinguishing this defect from a large secundum atrial septal defect (ASD) by echocardiography is often challenging. Accurate diagnosis of an inferior SVD and correct differentiation from a secundum ASD is essential for appropriate presurgical planning. Absence of the posterior rim in the parasternal short-axis views may serve as a useful clue in diagnosing inferior SVDs. We sought to determine the utility of using the presence or absence of a posterior atrial rim in the parasternal short-axis view to help distinguish an inferior SVD from a secundum ASD. This sign may help clinch the diagnosis when subcostal imaging is suboptimal.

Methods: We retrospectively reviewed transthoracic echocardiograms from 15 patients with a known surgical diagnosis of an inferior SVD between 2004 and 2015. The presence or absence of a posterior rim in the parasternal short-axis view was determined by two primary investigators. The posterior rim was also evaluated in 14 patients with a secundum ASD repair as controls. Echocardiograms were then reviewed blindly by attending-level echocardiographers and cardiology fellows in training. Diagnostic accuracy was assessed both with and without the use of the posterior rim criterion. Statistical analysis was used to determine the effect of using the rim criterion on inferior SVD diagnosis. We also reviewed all surgically diagnosed secundum ASDs that were incorrectly diagnosed as inferior SVD by preoperative imaging and determined whether use of the posterior rim criterion would have resulted in the correct diagnosis.

Results: The posterior rim was absent in all 15 patients with a surgical diagnosis of inferior SVD and present in all 14 patients with a secundum ASD. For all observers, there was a statistically significant increase in diagnostic accuracy of inferior SVDs with the use of the rim criterion ($P < .0001$). We noted that secundum ASDs with inferior extension also have persistent posterior rims. The rim criterion correctly classified all large secundum ASDs with inferior extension that were previously misdiagnosed by echocardiogram preoperatively.

Conclusions: Absence of the posterior rim ("bald" posterior wall) is a consistent finding in patients with an inferior SVD and distinguishes an inferior SVD from a large secundum ASD with inferior extension. Parasternal short-axis evaluation of the posterior atrial rim is a helpful tool for all levels of physician training in improving diagnostic accuracy for detecting inferior SVDs and in distinguishing them from secundum ASDs. (J Am Soc Echocardiogr 2017;30:209-15.)

Keywords: Inferior sinus venosus defect, Atrial septal defect, Echocardiogram

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Abbreviations**3D** = Three-dimensional**3DE** = Three-dimensional echocardiography**3D-TEE** = Three-dimensional transesophageal echocardiography**3D-TTE** = Three-dimensional transthoracic echocardiogram**ASD** = Atrial septal defect**AUC** = Area under the curve**CT** = Computed tomography**IVC** = Inferior vena cava**MRI** = Magnetic resonance imaging**ROC** = Receiver operating characteristic**SVD** = Sinus venosus defects**TEE** = Transesophageal echocardiography**TTE** = Transthoracic echocardiogram

Sinus venosus defects (SVD) are a rare form of congenital heart disease involving an interatrial communication outside the confines of the fossa ovalis. Although SVDs do not involve the atrial septum proper (i.e., the fossa ovalis and its surrounding muscular rim), they are placed in the same physiological category as atrial septal defects (ASD) and account for up to 11% of interatrial communications.¹⁻⁴ SVDs of the inferior vena cava (IVC) type, or inferior SVDs, involve a deficiency in posteroinferior aspects of the atrial septum, resulting in a communication between the right and left atrial chambers through the mouth of the IVC.^{2,5,6} An intact fossa ovalis and the IVC straddling over the defect are considered additional prerequisites for diagnosing inferior SVDs. Anomalous pulmonary venous drainage to the IVC is a common finding and has been

regarded by some investigators as an anatomical criterion for the diagnosis of inferior SVDs,^{2,5,6} whereas others have not included this feature as a diagnostic criterion¹ because it is not present consistently.

In contrast to the secundum type of ASD, inferior SVDs are not amenable to transcatheter device closure and thus require surgical repair. The presence of an inferior SVD may also influence the specific placement of the IVC cannula prior to cardiopulmonary bypass.¹ Previous studies have reported challenges with accurate preoperative diagnosis of inferior SVDs, with the defects often being mistaken for large secundum ASDs with inferior extension.⁵⁻⁷ While combination defects within the atria may be present, defining the posteroinferior rim anatomy itself has profound implications upon treatment strategy and overall hospital course. For example, Banka and colleagues found that misdiagnosis of inferior SVDs was associated with longer hospitalizations, a more complicated hospital course, and poorer technical outcomes.⁶

The importance of presurgical diagnostic precision has resulted in the use of multiple imaging modalities including transesophageal echocardiography (TEE) and magnetic resonance imaging (MRI).^{8,9} Additionally, many studies have described transthoracic echocardiographic (TTE) findings to help facilitate diagnosis of inferior SVDs and their distinction from secundum ASDs. However, in these studies, the focus was upon the absence of an IVC rim in the subcostal sagittal views.^{1-3,5,7} Frequently in children, the subcostal coronal and sagittal views and many “in-between” views are obtained, along with methodical sweeping along a stack of planes in each view. This allows the diagnosis of SVDs by creating a mental three-dimensional (3D) picture of the position of the defect with respect to the IVC. However, this method also requires optimal images, which may be lacking in some older children and adults. Moreover, in adult echocardiography laboratories, such multiplanar subcostal imaging of the atrial septum is performed much less frequently. Previous studies have suggested that the para-

sternal short-axis view may demonstrate a deficiency in the posterior atrial rim.¹⁻³

Our aim was to determine whether an absent posterior atrial rim in the parasternal short-axis view was a consistent and reliable finding in all patients with inferior SVDs. We hypothesize that the identification of a “bald atrial wall,” seen in the absence of a posterior atrial rim in a parasternal short-axis view, is a consistent feature found in inferior SVDs, which will improve diagnostic accuracy.

METHODS

We retrospectively reviewed the clinical data, echocardiography, and surgical notes from our surgical database for all patients between ages 0 and 18 years of age with a postoperative diagnosis of an inferior SVD at our institution over an 11-year time period, between January 2004 and February 2015. Subjects diagnosed with a secundum ASD who subsequently underwent surgical closure from the same time period were used as controls.

To be included in the study, patients must have had a full echocardiogram performed prior to surgical intervention. The minimal criterion for an inferior SVD defined for this study was an atrial communication originating at the mouth of the IVC with the IVC straddling the defect. The presence of anomalous pulmonary venous drainage was noted when present but not required to establish a diagnosis of inferior SVD. Our posterior rim criterion was defined as absence of a posterior atrial rim in the standard parasternal short-axis view (giving a “bald” appearance to the posterior atrial wall). This criterion was used to support the diagnosis of inferior SVD, while the presence of a posterior rim, even a rudimentary one, went against this diagnosis (Figure 1A–D). In some patients, a slightly modified parasternal short-axis view obtained by 20° counterclockwise rotation of the transducer was also available, as this view is frequently used in our institution to clarify the position of an ASD with respect to the IVC.

Echocardiograms were reviewed by two investigators (B.S.S., A.B.) to determine the presence or absence of a posterior rim. We then sought to determine whether this particular echocardiographic finding would facilitate the accurate diagnosis of inferior SVDs. Echocardiograms were reviewed blindly by three separate attending physicians who were more than 8 years past their fellowship training and were dedicated echocardiographers. Echocardiograms were also reviewed by two first-year cardiology fellows who were at the end of their first year of training (fellows 1 and 2) and three second-year fellows (fellows 3, 4, and 5). Lastly, we performed a reverse analysis, and all the patients with a surgical diagnosis of secundum ASD who were incorrectly diagnosed as having an inferior SVD in preoperative echocardiographic report were identified and the presence of the posterior rim was determined by the investigators of this study. The study was approved by the Institutional Review Board of the Children’s Hospital of Philadelphia (IRB no. 14-011648).

Statistics

Descriptive statistics of the demographic factors were reported for the two groups of patients separately. Percentage of accurate diagnosis was calculated and reported. Determination of diagnostic accuracy was done by performing a logistic regression analysis and generating receiver operating characteristic (ROC) curve, and the area under the curve (AUC) was calculated as a measure of discriminatory power of the test. The values used for the predictor were either inferior SVD or secundum ASD. McNemars test was used to determine statistical significance of using the posterior rim criterion

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