

Sinus Venosus versus Ostium Secundum Atrial Septal Defects: Their Diagnosis and Surgical Outcome

Hammood Naser Mohsin, Firas Sadeq Abdul Kareem

Ibn Al Nafees Cardiac Centre, Baghdad, Iraq

Abstract

Background: Diagnosis of sinus venosus atrial septal defects (ASDs) poses clinical difficulties than that of ostium secundum ASDs which may delay its diagnosis and treatment. Surgical repair of sinus venosus ASDs is more intricate in correlation with the repair of ostium secundum ASDs and conveys the risk of stenosis of the superior vena cava (SVC) or the rerouted pulmonary veins (PVs) and sinus node dysfunction (SND). **Objectives:** To compare diagnostic modalities and surgical outcome of sinus venosus ASDs to that of ostium secundum ASDs. **Patients and Methods:** Out of the total 72 patients in the age range of 1 year–46 years, 36 patients underwent surgical repair of sinus venosus ASDs and 36 patients underwent surgical repair of ostium secundum ASDs. Their data were collected and retrospectively studied from June 1, 2009, to October 1, 2016, at Ibn-Alnafees Teaching Hospital for cardiothoracic surgery in Baghdad, Iraq. We divided the patients into two groups: Group A including 36 patients who had sinus venosus ASDs and Group B including 36 patients who had ostium secundum ASDs. A comparison was made between the two groups according to preoperative variables, diagnostic modalities, intraoperative variables, and postoperative morbidity and mortality. **Results:** Transthoracic echocardiography (TTE) was the mainstay for the diagnosis of ASD secundum, while ASD venosus type needed more modalities for diagnosis. No operative death, postoperative bleeding, acute renal failure, SND, SVC stenosis, and PV stenosis were observed in any patient in the two groups. Residual shunt was not observed in any case in the two groups. **Conclusion:** TTE, transesophageal echocardiography, cardiac magnetic resonance imaging, or diagnostic catheterization may be needed for the diagnosis of sinus venosus ASD. Surgery is the mainstay of sinus venosus ASDs with low morbidity and no mortality, similar to ostium secundum ASDs.

Keywords: Anomalous pulmonary vein, idiopathic pulmonary hypertension, ostium secundum atrial septal defect, right atrium, single-patch procedure, sinoatrial node, sinus venosus atrial septal defect

INTRODUCTION

Ostium secundum atrial septal defects (ASDs) are the most widely recognized variation of ASD comprising roughly 75% of all ASDs where the imperfection is at the center portion of the atrial septum (fossa ovalis).^[1,2] Sinus venosus ASD (SVASD) is defined as a deficiency of the atrial septum at the insertion of the superior vena cava (SVC) or the inferior vena cava (IVC) into the right atrium (RA) (superior and inferior type SVASD, respectively). SVASD imperfections represent only 10% of atrial septal deformities.^[3] There are no less than at least one anomalous pulmonary vein(s) (PV) to the SVC or inferior vena cava or the RA.^[4] Transthoracic echocardiography (TTE) stays first line in indicative assessment; however, identification rates have been accounted for as low as 12%–44%.^[5,6] Transesophageal echocardiography (TEE) enhances demonstrative precision;

however, it may not distinguish ordinarily related partial anomalous pulmonary venous return (PAPVR).^[7] Cardiac magnetic resonance imaging (MRI) gives a noninvasive, exceptionally delicate and particular imaging methodology of SVASD.^[8] Electrocardiography (ECG)-gated heart computed tomography has proven to precisely recognize related PAPVR.^[9] Cardiac catheterization is typically not required in the preoperative appraisal of patients with sinus venosus atrial septal deformities.^[10] Careful amendment of SVASD

Address for correspondence: Dr. Firas Sadeq Abdul Kareem,
Ibn Al Nafees Cardiac Centre, Baghdad, Iraq.
E-mail: firmas_alkubaisy2004@yahoo.com

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is more intricate in correlation with the repair of ostium secundum ASDs and conveys the risk of stenosis of the SVC or the rerouted PVs and sinus node dysfunction (SND).^[11,12] Different surgical procedures have been used in order to avoid these complexities including the single-patch strategy which remains the method of decision for sinus venosus atrial septal imperfections with partial anomalous pulmonary venous connection step inside the RA or RA-SVC intersection; double-patch technique which limits the risk of SVC stenosis which, however, may result in sinoatrial (SA) node dysfunction by disrupting the SA node or the SA node artery; and Warden procedure which has picked up prevalence for avoiding both pulmonary venous and SVC obstruction as well as SA node dysfunction.^[13] Excellent surgical results with a mortality rate near 0% can be expected. This is particularly true in patients who undergo repair when younger than 15 years.^[14]

Aim of this study

To compare between SVASD and ASD secundum in relation to diagnostic modalities, surgical procedures, postoperative complications, and any associated anomalies

PATIENTS AND METHODS

Out of a total of 72 patients in the age range of 1 year–46 years, with a median age of 23 ± 7 years, 36 patients underwent surgical repair of SVASD (superior type) and 36 patients underwent surgical repair of ostium secundum ASD. Their data were collected from the outpatient clinic and retrospectively studied from June 1, 2009, to October 1, 2016, at Ibn Al Nafees Teaching Hospital for cardiothoracic surgery in Baghdad, Iraq. Most cases were referred from pediatric cardiology unit to cardiac surgery unit, and all patients were evaluated by pediatric cardiologist preoperatively and postoperatively. ECG, chest X-RAY and TTE was applied to all patients, TEE was done for 13 patients in group A, cardiac catheterization was needed in 11 patients in group A and 2 patients in group B. All patients with SVASD had associated anomalies; 35 patients had anomalous right superior PV, one patient had anomalous right superior PV and right middle vein, three patients had coexisting ostium secundum ASD, and two patients had valvular pulmonary stenosis. Surgery was performed using classical median sternotomy, total cardiopulmonary bypass, and antegrade cardioplegia in all patients. Intraoperative TEE was not employed in any patient. All cases were assessed postoperatively by TTE in the early postoperative period and before discharge. We divided the patients into two groups: Group A included 36 patients who had SVASD and Group B included 36 patients who had ostium secundum ASD. In this study, we excluded any case of ostium secundum ASD closed by transcatheter approach or where it is a part of more complex congenital cardiac disease. A comparison was made between the two groups according to preoperative variables (age, gender, body weight, body surface area, and presented feature), diagnostic modalities, intraoperative variables (aortic cross-clamp time and cardiopulmonary bypass time), intubation time, intensive care

unit stay, need of full inotropic support time, and postoperative morbidity and mortality. IBM SPSS Statistics for Windows, Version 22.0. Armonk, NY: IBM Corp. was used for authentic examination. The results were imparted by mean and standard deviation for tenacious components or with repeat and rate for total variables. Pearson's Chi-square test was used for assessing the relationship between variables. $P < 0.05$ was found ideal to be quantifiably immense.

RESULTS

In this study, the commonest age group in group A was between 30-39 years (44.44%), while in group B, the commonest age was between 1-9 years (47.22%). The mean age was 28.39 years in Group A and 18.2 years in Group B. Female: male ratio was 3:1 in Group A and 3.5:1 in Group B [Table 1]. There was no operative death in the two groups. Postoperative bleeding, acute renal failure, SND, SVC stenosis, and PV stenosis were not observed in any group. Residual shunt also was not observed in any group. The incidence of respiratory tract infection, pericardial effusion, and tachyarrhythmia was insignificantly higher in Group A than Group B [Table 2]. TTE was the diagnostic modality in 19 patients in Group A (52.77%) versus 36 patients in Group B (100%). TEE was necessitated in 13 patients in Group A (36.11%), while no such need

Table 1: Age and gender distribution of patients

Variable	Group A (%)	Group B (%)	Total (%)	P
Age (years)				
1-9	1 (2.78)	17 (47.22)	18 (25)	0.028
10-19	3 (8.33)	9 (25)	12 (16.67)	0.057
20-29	9 (25)	5 (13.89)	14 (19.45)	0.234
30-39	16 (44.44)	3 (8.33)	19 (26.38)	0.0005
40-46	7 (19.45)	2 (5.56)	9 (12.5)	0.075
Total	36 (100)	36 (100)	72 (100)	
Gender				
Female	27 (75)	28 (77.78)	55 (76.38)	0.779
Male	9 (25)	8 (22.22)	17 (23.62)	0.779
Total	36 (100)	36 (100)	72 (100)	

Table 2: Comparison between the two groups according to morbidity and mortality after surgery

Variable	Group A (%)	Group B (%)	Total (%)	P
Respiratory tract infection	4 (11.11)	3 (8.33)	7 (9.72)	0.689
Bleeding	0	0	0	1.00
Acute renal failure	0	0	0	1.00
SA node dysfunction	0	0	0	1.00
SVC stenosis	0	0	0	1.00
Pulmonary vein stenosis	0	0	0	1.00
Pericardial effusion	3 (8.33)	2 (5.55)	5 (6.94)	0.645
Tachyarrhythmia	2 (5.55)	1 (2.77)	3 (4.16)	0.555
Residual shunt	0	0	0	1.00
Mortality	0	0	0	1.00

SA: Sinoatrial, SVC: Superior vena cava

was observed in Group B. Cardiac catheterization was used in 11 patients in Group A (30.55%) versus 2 patients in Group B (5.55%) [Table 3]. Direct closure was made in seven patients in Group B (19.44%), while it was not used in any patient in Group A. Single-patch technique was the common procedure in both groups (94.45% in Group A vs. 80.56% in Group B). Double-patch technique was applied in two patients in Group A (5.55%), while no such procedure was used in Group B [Table 3]. Mean body weight and body surface area were higher in Group A than Group B [Table 4]. Mean aortic cross-clamp time and cardiopulmonary bypass time were longer in Group A than Group B (43.35 min vs. 23.89 min and 59.21 min vs. 42.46 min, respectively), and mean intubation time and intensive care unit stay were little longer in Group A than Group B [Table 4]. Exertion dyspnea was a common presented feature in Group A (33.33%) versus fatigue in Group B (52.77%). Preoperative incidence of tachyarrhythmia and pulmonary hypertension was higher in Group A than Group B [Table 5].

DISCUSSION

In this study, mean age was 28.39 years in Group A versus 18.2 years in Group B which indicates that early presentation of ostium secundum ASD and easy to diagnose by routine TTE versus SVASD. Because of the uncommon predominance and anatomical many-sided quality, diagnosing sinus venous ASD poses clinical difficulties, which may defer diagnosis and treatment.^[6] Female: male ratio was 3:1 in Group A versus 3.5:1 in Group B; the overall ratio was higher than that of other studies abroad where female: male ratio was 2:1, which may reflect the high percentage of females in our population. There was no operative death and the mortality rate was 0% in the two groups, which reflects the safety of surgery in both types of ASD. SND, SVC stenosis, and PV stenosis are the major complications which may be observed in SVASD.^[8-10] In our study, those complications were not observed in any patient in the two groups, which reflects the low morbidity in both groups postoperatively. Residual shunt was not observed in any case in the two groups, which reflects the feasibility of surgery to close those types of ASD. Other major complications such as postoperative bleeding and acute renal failure were not found in the two groups. Mean aortic cross-clamp time and cardiopulmonary bypass time were longer in the surgery of SVASD than that of ostium secundum ASD, which reflects that the surgery of SVASD was more technique dependent, but this did not lead to any adverse effect on outcome, where the mean intubation time and intensive care unit stay were approximately equal in two types of ASD. Moreover, there was no need of full inotropic support in the two types of ASD. Clinical presentations were variable; exertion dyspnea represented 33.33% in Group A, which was a common one, whereas fatigue was the common presentation in Group B (52.77%), which may reflect delay in the diagnosis of SVASD until dyspnea appeared. Pulmonary hypertension was observed in four patients (11.11%) in Group A (SVASD)

Table 3: Diagnostic modalities and surgical procedures used in the two groups

Diagnostic modality	Group A (%)	Group B (%)	Total (%)	P
TTE	19 (52.77)	36 (100)	72 (100)	0.234
TEE	13 (36.11)	0	13 (18.06)	0.003
Cardiac MRI	0	0	0	1.00
ECG-gated heart CT	0	0	0	1.00
Cardiac catheterization	11 (30.55)	2 (5.56)	13 (18.06)	0.005
Surgical procedure				
Direct closure	0	7 (19.44)	7 (9.72)	0.0054
Single patch	34 (94.45)	29 (80.56)	63 (87.5)	0.075
Double patch	2 (5.55)	0	2 (2.78)	0.152
Warden procedure	0	0	0	1.00

ECG: Electrocardiography, CT: Computed tomography, TTE: Transthoracic echocardiography, TEE: Transesophageal echocardiography, MRI: Magnetic resonance imaging

Table 4: Mean values of variables of Group A and Group B

Variable	Group A	Group B	Total	P
Body weight	36.7	12.8	24.75	0.021
Body surface area	1.06	0.84	0.95	0.085
Aortic cross-clamp time (min)	43.35	23.89	33.62	0.342
Cardiopulmonary bypass time (min)	59.21	42.46	50.83	0.624
Intubation time (h)	4.98	4.37	4.67	0.986
Intensive care unit stay (day)	2	1.8	1.9	0.875
Need of full inotropic support time (day)	0	0	0	1.00

Table 5: Major clinical presenting features of patients

Variable	Group A (%)	Group B (%)	Total (%)	P
Exertion dyspnea	12 (33.33)	7 (19.45)	19 (26.39)	0.180
Fatigue	6 (16.67)	19 (52.78)	25 (34.72)	0.001
Repeated chest infection	5 (13.89)	3 (8.33)	8 (11.11)	0.453
Arrhythmia	3 (8.33)	2 (5.56)	5 (6.95)	0.645
Paradoxical embolism	1 (2.78)	0	1 (1.39)	0.315
Pulmonary hypertension	4 (11.11)	3 (8.33)	7 (9.72)	0.689
Asymptomatic	5 (13.89)	2 (5.55)	7 (9.72)	0.234
Total	36 (100)	36 (100)	72 (100)	

where three of them were previously diagnosed as idiopathic pulmonary hypertension versus no case of ostium secundum ASD was previously diagnosed as idiopathic pulmonary hypertension which reflect much difficulty in the diagnosis of SVASD than ostium secundum ASD. TTE was applied to all patients which was a diagnostic tool in all patients in Group B (100%) where only two patients (aged 40 years and above) needed coronary angiography to rule out ischemic heart disease, whereas TTE was diagnostic in 19 patients (52.77%) in Group A where 13 patients (36.11%) required TEE and 4 patients required cardiac catheterization to confirm the diagnosis, which reflect difficulty of diagnosis SVASD which

requires more advanced diagnostic modalities. In our study, the frequency of TTE usage as the only used diagnostic modality in SVASD was higher as compared to other studies in worlds were only (44%) and this reflects that many patients may miss due to underestimation their conditions because dependability on it and not use other diagnostic images.^[5,6] TEE was applied in 13 patients only (36.11%). Other international studies regard this type of diagnosis as a standard method for diagnosis due to posterior position of defect and associated anomalous PV(s), which reflects that our experience in TEE is still far away from that abroad.^[7] Cardiac catheterization was applied to 11 patients (30.55%) in Group A, of which seven were in the age group of 40–46 years. This was applied to exclude any associated coronary artery disease; other four patients applied to confirm the diagnosis which do not accept to do this invasive procedure for diagnosis and reflect underuse of other noninvasive diagnostic modalities which may be due to lack of experience like TEE or unavailability like cardiac MRI.^[8] Direct closure was applied to seven patients (19.44%) in Group B versus no case in Group A, which reflects the complexity of SVASD where all patients had associated anomalous PV (s) (100%), right superior PV was the common (97.22%) which necessitate the usage of a patch as a baffle and proximity to SVC which required patch usage to prevent SVC obstruction.^[11,12] Single-patch procedure was applied in 34 patients (94.45%) in Group A versus 29 patients (80.56%) in Group B. This procedure was common to both groups, so the majority of SVASD and ostium secundum ASDs were closed by the same procedure. Double-patch procedure was required in two patients (5.55%) in Group A (SVASD), whereas there was no such requirement of this procedure in Group B, which reflects the complexity of SVASD. The SVASD technique was used to prevent SVC or PVs stenosis. The study limitation is the lack of long-term follow-up of both groups.

CONCLUSION

Diagnosis of SVASD is more difficult than that of ostium secundum ASD, which is usually confirmed by TTE, while SVASD needs more advanced diagnostic modalities like TEE; hence, more experience is needed in TEE as it a standard diagnostic modality abroad.

Noninvasive advanced cardiac imaging such as cardiac MRI should be used to rule out the need for invasive diagnostic modality (cardiac catheterization) in diagnosing SVASD.

All patients diagnosed as idiopathic pulmonary hypertension must be checked in specialized cardiac centers to exclude SVASD.

Surgery is the mainstay of treatment for SVASD with low morbidity and mortality rates, similar to ostium secundum ASD.

Single-patch procedure is the principal surgical procedure in SVASD with anomalous PV(s) drain into the RA–SVC intersection.

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Conflicts of interest

There are no conflicts of interest.

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