

An adult patient with previously undiagnosed sinus venosus atrial septal defect presenting with brain abscess: a case report

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Received 31 March 2021; first decision 20 April 2021; accepted 9 June 2021

Background

Brain abscess is a common complication in children with cyanotic congenital heart disease. The presence of an underlying acyanotic congenital heart disease is usually not suspected in an adult patient presenting with brain abscess.

Case summary

A 51-year-old male patient with no known co-morbidities came with complaints of recent onset right lower limb weakness needing support while walking and on evaluation was found to have brain abscess. He underwent robotic endoscope assisted endoport excision of the brain abscess. Two-dimensional transthoracic echocardiography showed right atrial and right ventricular dilatation with mild low-pressure tricuspid regurgitation. Transoesophageal echocardiography (TOE) revealed sinus venosus atrial septal defect (ASD) with left-to-right shunt with the right upper pulmonary vein draining into superior vena cava. Contrast echocardiography revealed a small transient right-to-left shunt. He has been advised to undergo elective surgical closure of ASD with partial anomalous pulmonary venous connection repair.

Discussion

Right-to-left shunting in ASDs can occur in the early systole even in the absence of raised pressures in the right side of the heart, even when the predominant shunt is left to right, but the magnitude of such a shunt is small and transient and is easily missed. Contrast echocardiography and TOE should be done as a part of evaluation of patients presenting with brain abscess.

Keywords

Case report • Atrial septal defect • Sinus venosus atrial septal defect • Sinus venosus atrial septal defect with partial anomalous pulmonary venous connection • Brain abscess

Learning points

- Brain abscess can occasionally be a presenting manifestation of acyanotic congenital heart disease with predominant left to right shunt.
- Right to left shunting in atrial septal defects can occur in the early systole even in the absence of raised pressures in the right side of the heart.
- A complete cardiac evaluation including contrast echo and transoesophageal echocardiography should be done as a part of evaluation of cerebral abscess.

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Handling Editor: Sylvia Krupickova

Peer-reviewers: Filippo Puricelli; Mohammed Al-Hijji

Compliance Editor: Daniel Tardo

Supplementary Material Editor: Mariame Chakir

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Introduction

Brain abscess is one of the serious complications of cyanotic congenital heart disease.¹ It most commonly occurs in children and adolescents with a peak incidence at 4–7 years of age.¹ Sinus venous atrial septal defect (SVASD) are located close to the SVC, and usually above the fossa ovalis, and it constitutes about 2–3% of all interatrial communications.^{2,3} These defects do not have a superior margin.³ It is frequently (80–90%) associated with anomalous pulmonary venous return from the right lung, usually of the right upper or middle lobe pulmonary veins to either the SVC or right atrium.² In size, the sinus venous defect may vary from small (and clinically unrecognizable) to non-restrictive.² The superior vena caval vein tends to override the defect, allowing for a biatrial superior caval connection.² In contrast

to other types of atrial septal defects (ASDs), SVASD is not known to close spontaneously.⁴ In a study in 115 patients with SVASD by Josh *et al.*, the usual manifestation was NYHA (New York Heart Association) functional class III or IV dyspnoea that was present in 20% of the patients. Apart from dyspnoea, the following symptoms and history were reported: palpitations (31%), angina (19%), history of congestive heart failure (9%), and history of stroke (3%).⁵ Diagnosis of SVASD on transthoracic echocardiography (TTE) is challenging and often necessitates the complementary use of more advanced studies, specifically transoesophageal echocardiography (TOE), cardiac magnetic resonance imaging (MRI), cardiac computed tomography, and rarely, cardiac catheterization.⁶ We are reporting a case who presented with central nervous system manifestations due to brain abscess and on evaluation was found to have sinus venous ASD.

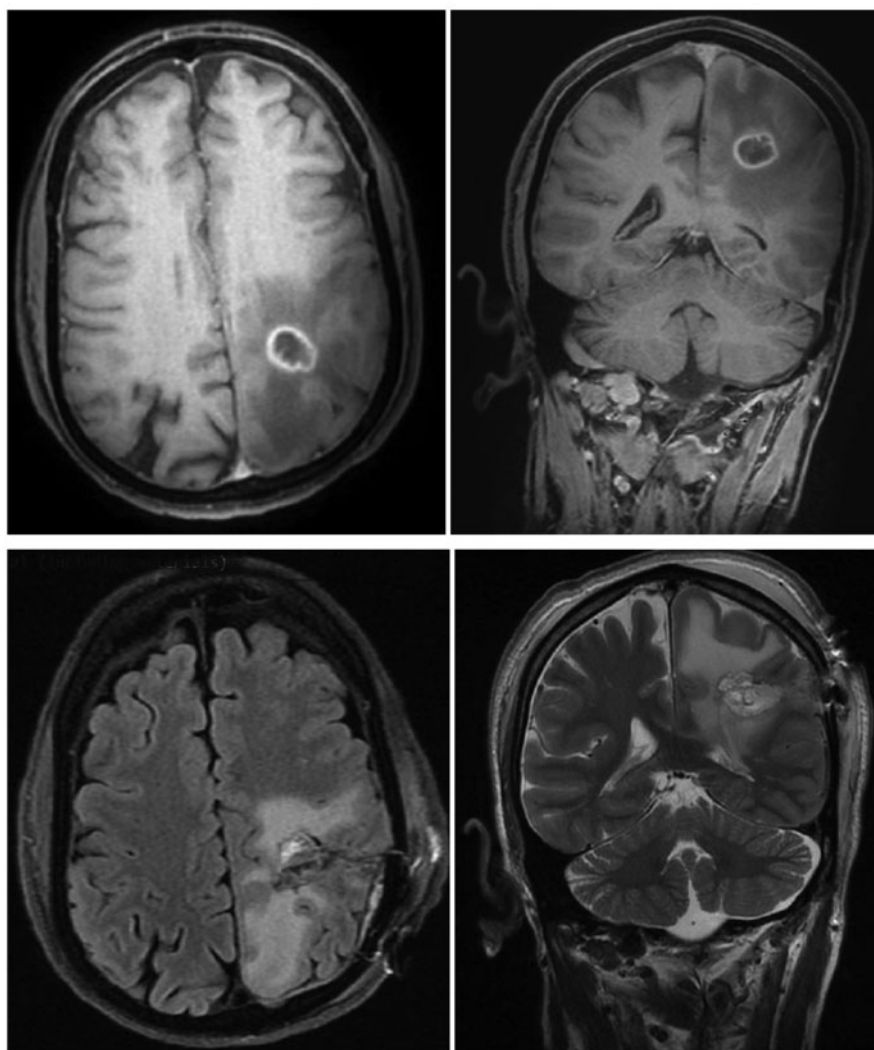
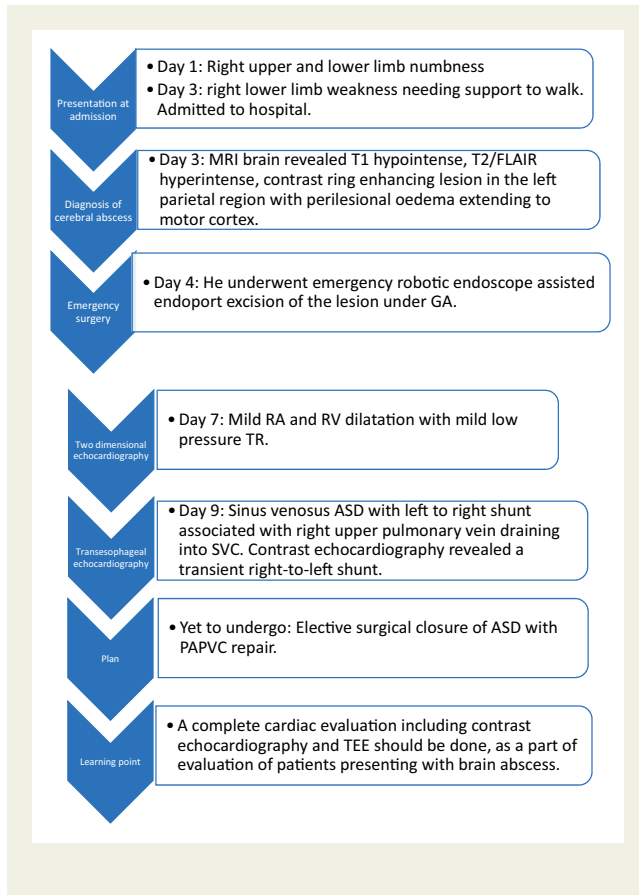


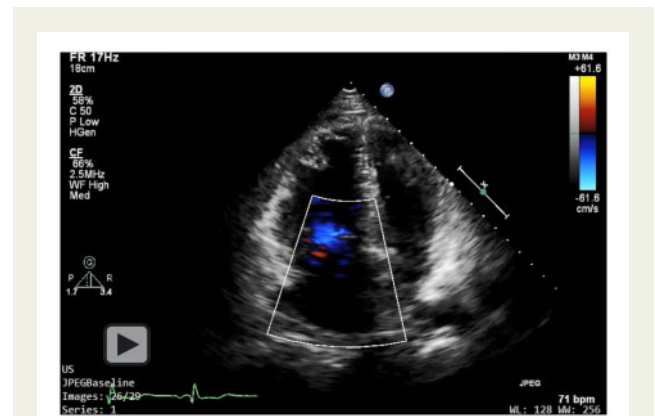
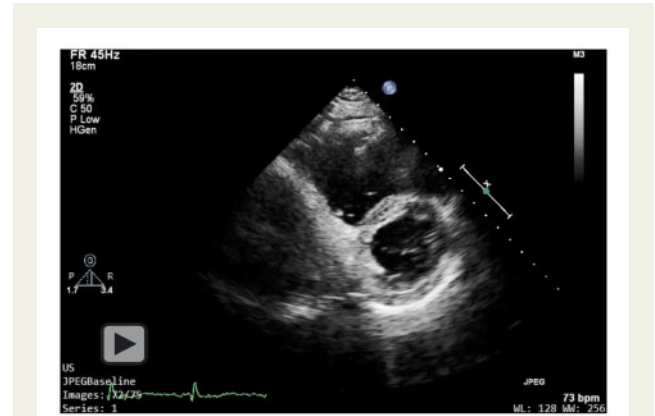
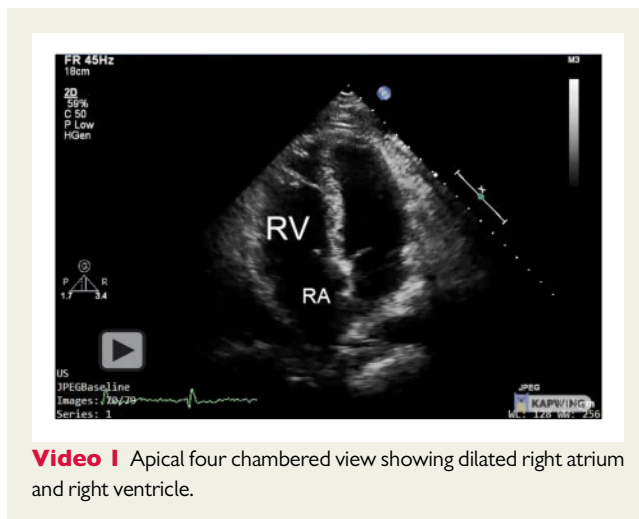
Figure 1 Above (right and left): T1-weighted contrast magnetic resonance imaging of brain showing ring enhancing lesion seen in left parietal lobe with surrounding oedema. Below: T2 FLAIR (below left) and T2-weighted (below right) magnetic resonance imaging images of brain showing post-operative changes in the left parietal lobe with areas of gradient blooming, suggestive of haemorrhage. Surrounding oedema in the left parietal and frontal lobes. Bifrontal pneumocephalus seen.

Timeline



Case presentation

A 51-year-old male patient with no known co-morbidities came with complaints of recent onset right upper and lower limb numbness and right lower limb weakness needing support while walking. He was



conscious and oriented, obeying commands, afebrile with stable vital signs on admission. His bilateral pupils were 2 mm reactive to light. His white blood counts were 12 470 cells/ μ L with 84.8% neutrophils and was initiated empirically on Ceftriaxone 2 g intravenous (IV) q8h and Amikacin 500 mg IV q12h. His MRI of the brain revealed a brain abscess in the left parietal region with perilesional oedema extending to motor cortex. There were no other infective foci evident on the MRI, hence any direct diffusion from a contiguous cranial infection site was excluded. He underwent emergency robotic endoscope assisted endoport excision of the lesion (Figure 1). Histopathology of the excised lesion showed glial tissue with dense inflammatory infiltrate comprised of lymphocytes, plasma cells, and histiocytes with focal suppurative inflammation and necrosis. Culture of the aspirate revealed moderate growth of pansensitive streptococcus species and was continued on the antibiotics for 2 weeks. Blood cultures were negative and dental work-up did not reveal any foci of infection. His human immunodeficiency virus status was negative and there was no history of IV drug use. Computed tomography scan of the chest and abdomen did not reveal any foci of infection. Levetiracetam 500 mg

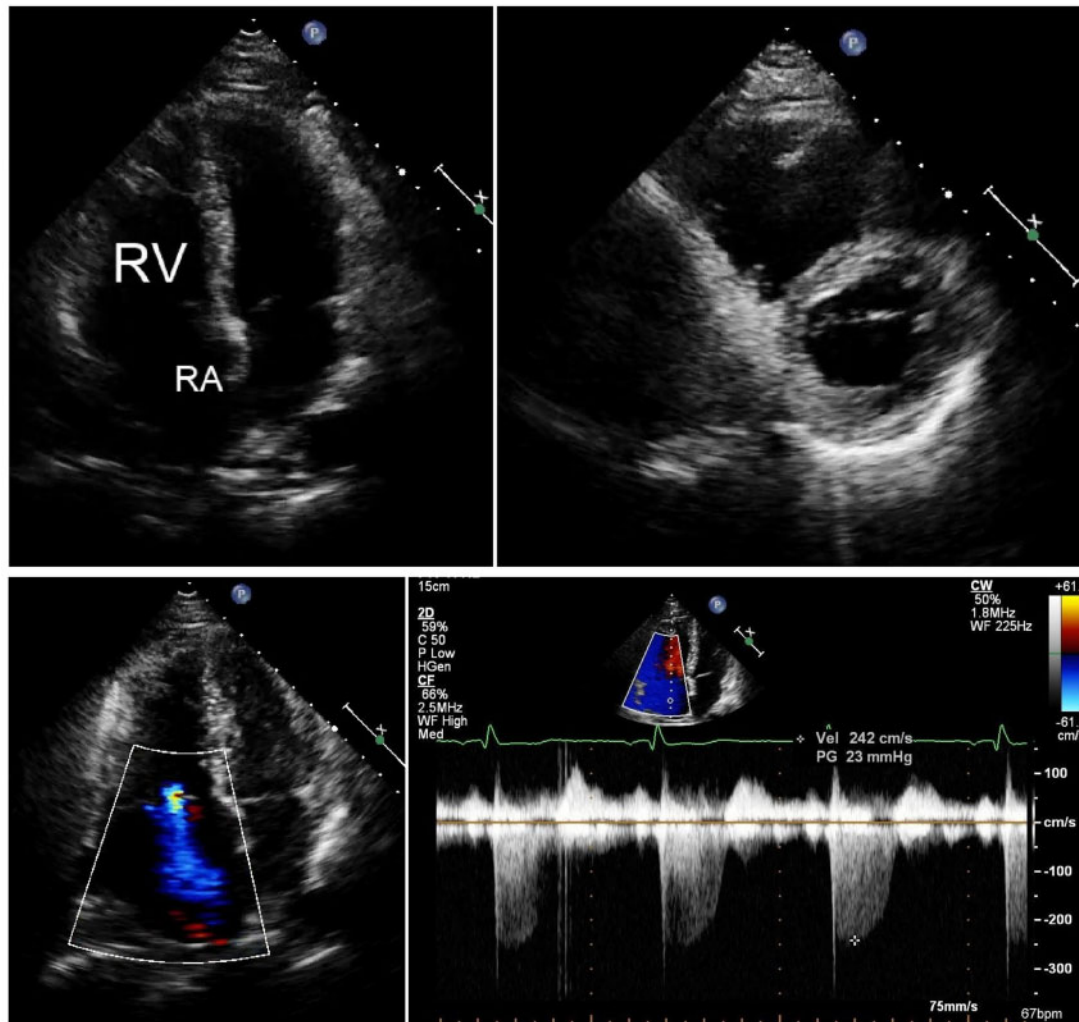


Figure 2 Above left: Apical four chambered view showing dilated right atrium and right ventricle. Above right: Parasternal short-axis view at the mitral valve level showing dilated right ventricle. Below left: Colour Doppler across the tricuspid valve showing mild tricuspid regurgitation. Below right: Continuous wave Doppler across the tricuspid valve showing a pressure gradient of 23 mmHg. RA, right atrium; RV, right ventricle.

intravenously q8h was initiated for seizure prophylaxis and was continued on tablet levetiracetam 500 mg q12h after discharge. As a part of routine evaluation to find out the source of abscess, cardiology consultation was sought. Two-dimensional TTE showed right atrial (RA) and right ventricular (RV) dilatation with mild low-pressure tricuspid regurgitation (Videos 1–3, Figure 2) and hence it was decided to further evaluate the patient with TOE. TOE revealed SVASD with left to right shunt associated with right upper pulmonary vein draining into SVC (Supplementary material online, Video S1, Figures 3 and 4). Contrast echocardiography revealed the presence of a small transient right-to-left shunt (Supplementary material online, Video S2, Figure 5). Other pulmonary veins were seen draining into left atrium. There was no vegetation. The patient was advised elective surgical closure of ASD with partial anomalous pulmonary venous connection repair without which the chances of recurrence is high.

Discussion

Brain abscess is focal intracerebral collection of pus that results from contiguous infection or haematogenous microbial spread and is a frequent manifestation of cyanotic congenital heart disease with right to left shunt.¹ Since ASD patients have left-to-right shunt, haematogenous abscess is uncommon in these patients. In adults with ASD, the onset of pulmonary hypertension provides right-to-left shunt predisposing to paradoxical embolism and brain abscess.² However, in our patient, pulmonary pressures appear normal at echocardiographic assessment. Right to left shunting in ASDs can occur in the early systole even in the absence of raised pressures in the right side of the heart, even when the predominant shunt is left to right, but the magnitude of such a shunt is small and transient and is easily missed⁷ and such a shunt was demonstrated in our patient on contrast echocardiogram (Supplementary material online, Video S2, Figure 5). Hence this can

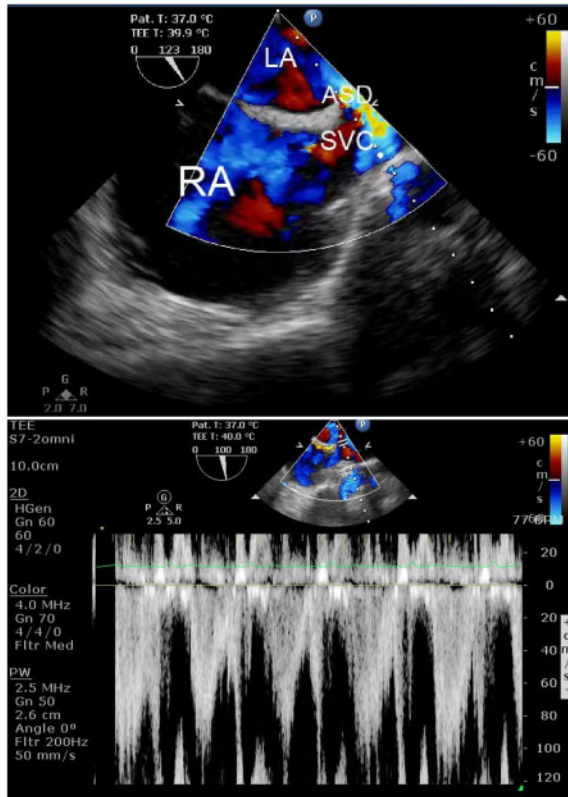


Figure 3 Above: Transoesophageal echocardiography showing sinus venosus atrial septal defect with left to right shunt. ASD, atrial septal defect; LA, left atrium; RA, right atrium; SVC, superior vena cava. Below: Pulse wave Doppler across the atrial septal defect showing left to right shunt.

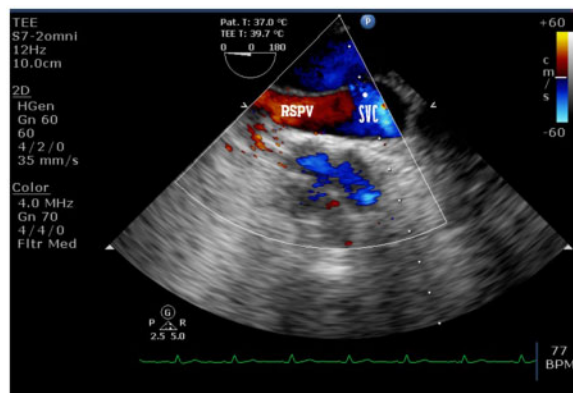


Figure 4 Short-axis oesophageal view of the superior vena cava at the level of the right pulmonary artery. Right superior pulmonary vein can be seen entering into medial aspect of the superior vena cava. RSPV, right superior pulmonary vein; SVC, superior vena cava.

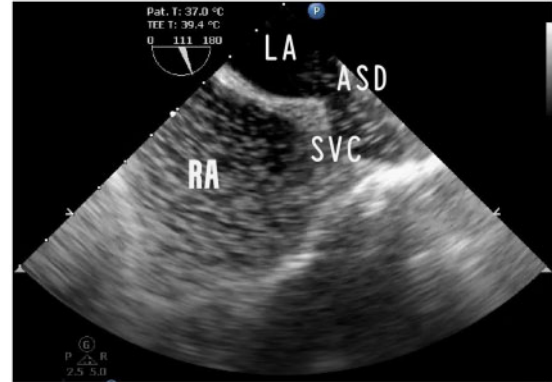


Figure 5 A small transient right-to-left shunt is evident by the movement of the bubble contrast from right atrium to left atrium almost immediately. ASD, atrial septal defect; LA, left atrium; RA, right atrium; SVC, superior vena cava.

justify the occurrence of cerebral abscess in our patient in spite of the presence of normal right heart pressures and predominant left to right shunt on echocardiogram. Hence a complete cardiac evaluation including contrast echocardiography and TOE should be done, especially if the suspicion is high as in our case where TTE revealed mild RA/RV dilatation, as a part of evaluation of patients presenting with brain abscess.

Lead author biography



Dr Navin Mathew received his MBBS degree in 1996 from Calicut University after his training from Trichur Medical College. Subsequently, he got training from the Stanley Medical College, Chennai and received his MD (Internal Medicine) in 2001. He joined the prestigious All India

Institute of Medical Sciences in December 2002 for his cardiology training and was awarded the Jagesh Lal Kapila medal for being the best post graduate in AIIMS cardiology 2005. Since 2006, Dr Navin is in the Department of Cardiology at the Amrita Institute of Medical Sciences and is a valuable member of the heart team.

Supplementary material

[Supplementary material](#) is available at *European Heart Journal - Case Reports* online.

Slide sets: A fully edited slide set detailing this case and suitable for local presentation is available online as [Supplementary data](#).

Consent: The authors confirm that written consent for submission and publication of this case report including images and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: None declared.

Funding: None declared.

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