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Editorial Board Member of *World Journal of Cardiology*, Akshyaya Pradhan, MD, DM, FACC, FSCAI, FESC, FAPSIC, Professor (Jr), Department of Cardiology, King George's Medical University, Lucknow, Uttar Pradesh 226003, India. akshyaya33@gmail.com

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CASE REPORT

Unroofed coronary sinus, left-sided superior vena cava and mitral insufficiency: A case report and review of the literature

Fouad Bitar, Ziad Bulbul, Yehya Jassar, Rana Zareef, Jennifer Abboud, Mariam Arabi, Fadi Fouad Bitar

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Fouad Bitar, Ziad Bulbul, Rana Zareef, Jennifer Abboud, Mariam Arabi, Fadi Fouad Bitar, Department of Pediatric and Adolescent, American University of Beirut Medical Center, Beirut 1103, Lebanon

Yehya Jassar, Department of Surgery, American University of Beirut Medical Center, Beirut 1103, Lebanon

Corresponding author: Ziad Bulbul, MD, Doctor, Department of Pediatric and Adolescent, American University of Beirut Medical Center, Beirut-Hamra-Cairo Street, Beirut 1103, Lebanon. zb21@aub.edu.lb

Abstract

BACKGROUND

Unroofed coronary sinus (UCS) is a rare subtype of atrial septal defect. It is frequently associated with a persistent left superior vena cava and is often part of a more intricate cardiac malformation.

CASE SUMMARY

This report describes a rare case of an adolescent patient with UCS featuring atrial situs solitus, absence of the right superior vena cava and a persistent left superior vena cava draining into the left atrium consistent with total unroofing of the coronary sinus. This was associated with concurrent severe mitral insufficiency secondary to redundant and prolapsing leaflets, and a substantial left-to-right shunt across the coronary sinus orifice. A comprehensive examination of the existing literature is included, shedding light on the diagnostic challenges of UCS and describing the available surgical options within the context of mitral valve surgery.

CONCLUSION

UCS is a complex condition requiring careful consideration of associated anomalies and a tailored surgical approach.

Key Words: Unroofed coronary sinus; Mitral insufficiency; Single left superior vena cava; Surgical options; Absent right superior vena cava; Case report

Core Tip: In this report, we present an exceedingly rare case of a teenager with an unroofed coronary sinus and a single persistent left superior vena cava in conjugation with severe mitral regurgitation secondary to redundant and prolapsing leaflets, in the absence of other associated cardiac anomalies. This peculiar condition was detected by echocardiography and corrected by appropriate surgical intervention.

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INTRODUCTION

Unroofed coronary sinus (UCS), also known as coronary sinus (CS) septal defect, denotes a particularly rare and uncommon subtype of atrial septal defects, accounting for less than 1% of all cases within this category[1]. It arises from the incomplete development of the left atrial venous folds during embryogenesis and was first elucidated by Raghib and colleagues in 1965[2]. This anomaly manifests as a communication between the left atrium and the CS, secondary to partial or complete absence of the CS roof, resulting in a left to right shunt.

UCS is an atypical atrial septal defect as it is characterized by interatrial shunting across a normal structure, the CS ostium[3,4]. It often co-exists with a persistent left superior vena cava (LSVC) and other concomitant cardiac abnormalities, further complicating its detection[3,4]. Isolated cases of CS-to-left atrial fenestration are exceedingly rare, increasing the intricacy of the diagnostic process[3-5]. Besides, the presence of a LSVC in the absence of the right superior vena cava (RSVC) is extremely uncommon with an estimated incidence of less than 0.13%[3]. While the congenital cardiac defects associated with absent RSVC show a wide spectrum[3,6], the association of absent RSVC, persistent LSVC and UCS, in the absence of congenital heart disease is immensely rare.

In this report, we present a unique case of absent RSVC in a visceroatrial situs solitus, persistent LSVC draining into UCS, associated with severe mitral insufficiency in the absence of atrioventricular septal defect. Furthermore, we present a comprehensive examination of the existing literature, tackling the diagnostic hurdles and exploring surgical interventions, with a particular emphasis on managing mitral valve pathology simultaneously.

CASE PRESENTATION

Chief complaints

A 19-year-old female was referred to our Children's Heart Center with the diagnosis of an atrial septal defect for further evaluation and treatment.

History of present illness

The patient had minimal exercise intolerance and assumed a sedentary lifestyle. She had normal growth and development. Notably, she was known to have congenital heart disease since early childhood, yet the family did not have documented records.

History of past illness

Besides her congenital heart disease, she did not have a significant medical history.

Personal and family history

She has no family history of congenital heart disease.

Physical examination

On physical examination, her vital signs were normal except for an oxygen saturation of 95%. She had a grade 2/6 systolic murmur at the apex and another grade 2/6 systolic ejection murmur at the left upper sternal border with fixed splitting of the second heart sound.

Laboratory examinations

An electrocardiogram (ECG) revealed a normal sinus rhythm and QRS frontal axis, with a wide P wave suggestive of left atrial enlargement (Figure 1).

Imaging examinations

A chest x-ray was significant for an enlarged cardiac silhouette with prominent right atrium and increased vasculature suggestive of congestion. An echocardiogram (Figure 2) showed right atrium and ventricle dilation, with preserved right

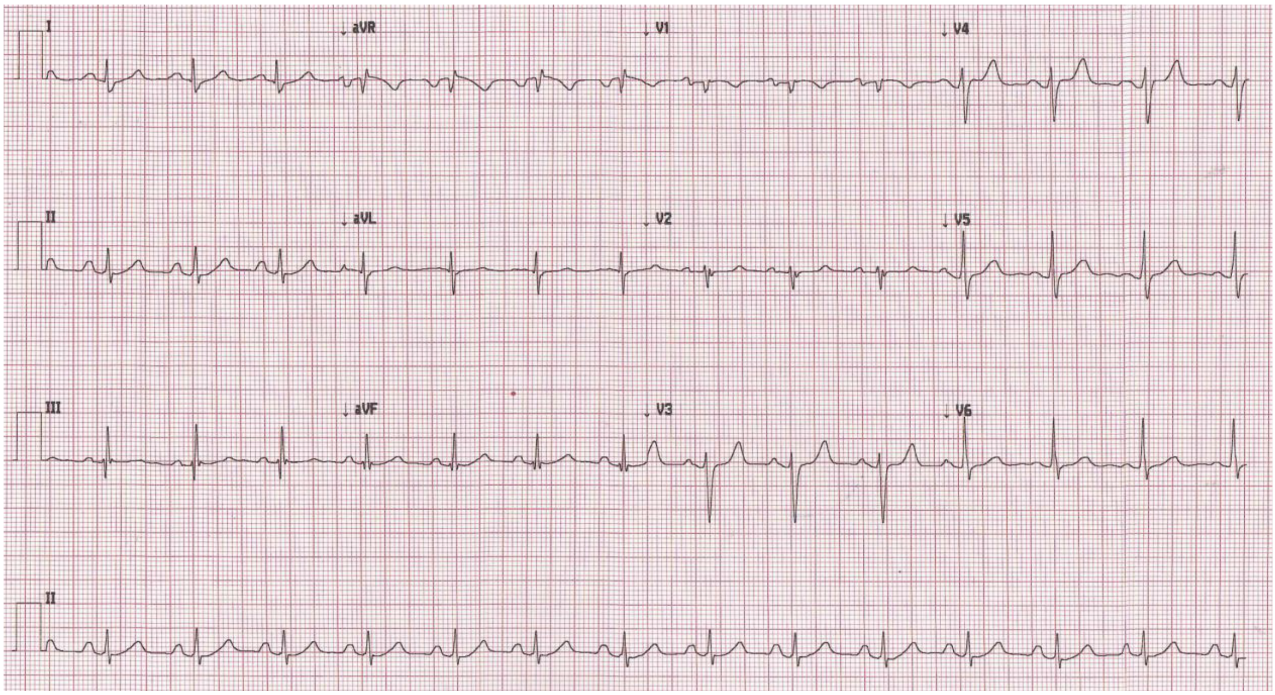

Figure 1 Pre-operative electrocardiogram. It shows normal sinus rhythm with normal P wave axis and interatrial conduction delay.

ventricular systolic function and absent RSVC. There was a right-sided brachiocephalic vein draining into a LSVC. Additionally, the ostium of the CS appeared significantly dilated with exuberant flow by color Doppler. The superior vena cava-CS continuity was interrupted consistent with UCS, while the LSVC was visualized to drain directly in the roof of the left atrium between the base of the left atrial appendage and the left upper pulmonary vein. Notably, the mitral valve leaflets were thickened, myxomatous and prolapsing resulting in multiple jets of severe mitral regurgitation. The mitral valve was bifoliate without a cleft, and the atrioventricular septum was intact without a primum atrial septal defect. Moderate tricuspid regurgitation was present and Doppler interrogation estimated the right ventricular systolic pressure to be around 30 mmHg. Contrast echocardiography, performed by injecting agitated saline through the antecubital vein of the left arm, showed sequential opacification of the LSVC, the left atrium, and the left ventricle. Concomitant with the opacification of the left ventricle, the right atrium and right ventricle opacified through the orifice of the CS (Figure 3). The inferior vena cava had a normal connection to the right atrium, and there was no evidence of a hemizygous connection. Pulmonary veins drained into the left atrium.

Subsequent computed tomography (CT) scan findings corroborated the echocardiographic diagnosis, confirming the presence of a single LSVC draining into the roof of the left atrium and complete unroofing of the CS. The ostium of the CS was dilated and served as the connection between the right atrium and the left atrium (Figure 4).

FINAL DIAGNOSIS

Workup confirmed the diagnosis of absent RSVC, persistent LSVC draining into the left atrium, and a completely UCS, in association with severe mitral insufficiency.

TREATMENT

The patient underwent surgical intervention as follows:

After median sternotomy, cardiopulmonary bypass was established *via* bi-caval venous and aortic cannulation. Upon inspection, the RSVC was absent and a persistent LSCV was noted. Antegrade cardioplegia was used. The right and left atrium were opened. Inspection of the right atrial cavity revealed a dilated and significantly enlarged CS ostium. Inspection of the left atrium showed the LSVC draining into the roof of the left atrium medial and superior to the orifices of the left pulmonary veins, and the base of the left atrial appendage was located just anterior and medial to the entrance site.

The repair of the LSVC to the left atrium was carried out using the repositioning of the atrial septum technique; a pericardial patch was sutured to the posterior rim of the LSVC using a continuous 5-0 polypropylene suture. The suture line was continued along the rim of the atrial septum and rim of the CS ostium where the caval orifice was positioned on the right side of this septum.

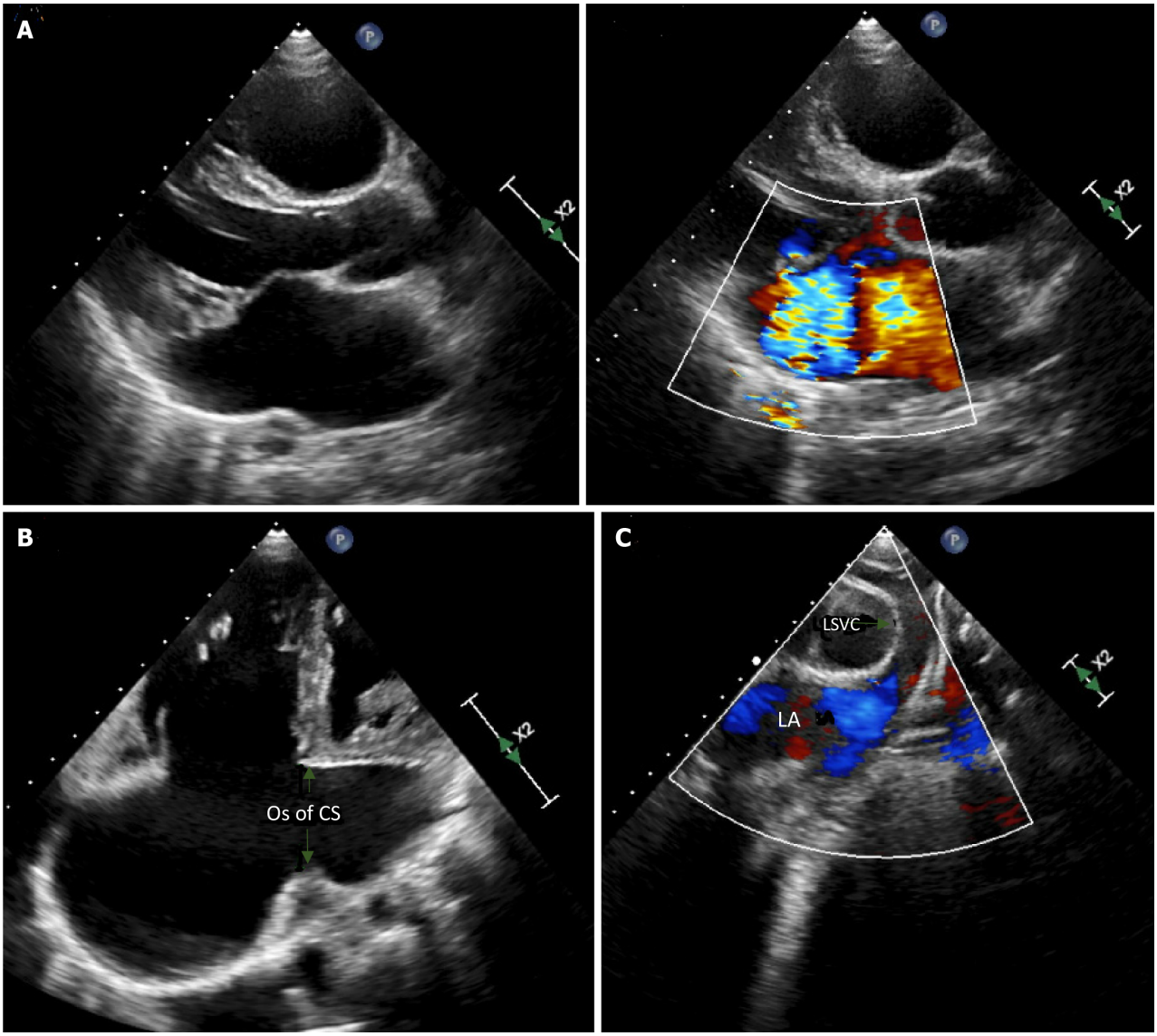

Figure 2 Echocardiographic imaging of the mitral valve. A: A parasternal long axis view of the mitral valve showing myxomatous prolapsing leaflets and severe mitral regurgitation; B: A modified 4 chamber view, with posterior angulation showing the dilated ostium of the coronary sinus; C: Modified supra coronal cut from the supra-sternal window showing a left superior vena cava draining into the left atrium. OS: Ostium; CS: Coronary sinus; LSVC: Left superior vena cava; LA: Left atrium.

Examination of the mitral valve confirmed the absence of a cleft and the severe prolapse of the anterior and posterior leaflets. The mitral valve repair was performed using the 4-chord technique with annuloplasty as described by Chemtob *et al*[7].

OUTCOME AND FOLLOW-UP

Post-operatively, a repeat CT scan and a LSVC angiogram revealed a patent intra-atrial baffle without any significant residual shunt (Figure 5).

DISCUSSION

This report describes a unique case of absent RSVC in a visceroatrial situs solitus, persistent LSVC draining into the left atrium, and a completely UCS, in association with severe mitral insufficiency in the absence of atrioventricular septal defect (AVSD). UCS is a rare condition often associated with other congenital anomalies. It exhibits various types and classifications, such as the Kirklin and Barratt-Boyes classification which utilizes the extent of the unroofing and the presence or absence of the LSVC[8].

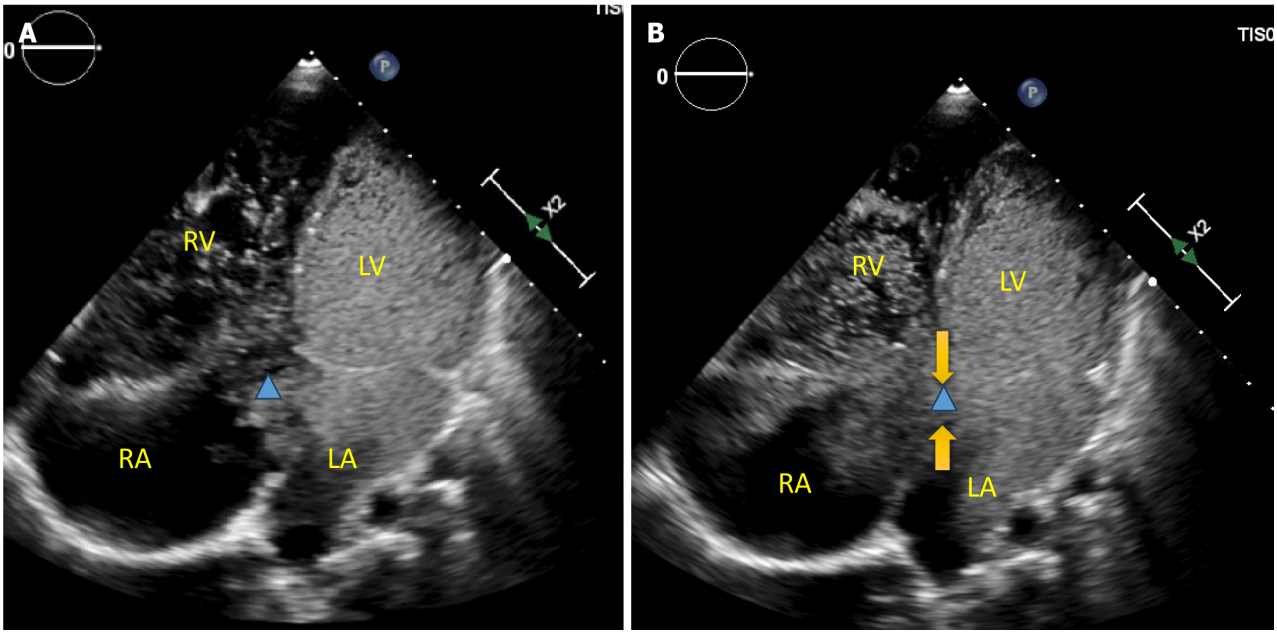

Figure 3 Agitated saline contrast injection in the left antecubital vein. A: Views taken from the 4-chamber window. Note the contrast filling the left atrium and the left ventricle before crossing the dilated ostium of the unroofed coronary sinus (triangle); B: Arrows pointing to the edges of the coronary sinus ostium. RA: Right atrium; LA: Left atrium; RV: Right ventricle; LV: Left ventricle.

The combination of an UCS, absent RSVC and a persistent LSVC is extremely rare in patients without heterotaxia syndrome. The rare obliteration of the right anterior cardinal vein and persistence of the left one during embryological development results in the absent RSVC with persistent LSVC[9]; if occurred concomitantly with failure of the formation of the left atrio-venous fold results in UCS[2]. Specific genes implicated in the association between UCS and persistence of LSVC have not been yet identified. However, both defects are linked to abnormal development of the venous system during fetal life. These anomalies are also frequently associated with genetic syndromes such as Noonan syndrome and Holt-Oram syndrome suggesting a potential genetic contribution.

Our patient had visceral and atrial situs solitus as confirmed by the P wave morphology and axis on the ECG, the drainage of the supra-hepatic portion of the inferior vena cava by imaging and ultimately the usual disposition of the atrial appendages upon direct inspection by the surgeon.

Bertram *et al*[3] reported on 121 cases, with absent RSVC, and normal cardiac lateralization, UCS was found in only seven patients: One with a ventricular septal defect, another with tetralogy of Fallot, a third with double outlet right ventricle and the remaining four did not exhibit associated congenital heart disease[10]. Martinez and his group reported an additional 12 cases, none had an UCS[7]. Doksöz *et al*[11] reported a child with absent RSVC, persistent LSVC which drained to the UCS, in association with AVSD and cor-triatriatum sinister. Kumar and his group reported a patient with absent RSVC, persistent LSVC draining into the UCS, a common atrium and an AVSD[12]. Partial UCS syndrome with persistent LSVC, absent RSVC and right-sided pericardial defect was also described in a pediatric patient[13].

UCS is rarely associated with mitral valve diseases, with only sporadic reports of mitral insufficiency in adult patients or in association with AVSD, where the regurgitation is usually through a cleft[14-17]. Mitral stenosis was reported once in the literature[18]. Detailed information on those cases is reported in Table 1.

In our patient, the presence of severe mitral insufficiency was associated with neither congenital heart disease, nor a cleft. Upon direct inspection, the mitral valve was free of a cleft, and the cause of the regurgitation was due to severe prolapse of both leaflets with elongated chords. Of interest, those changes could not have been attributed to age as is the case in adults with Barlow's disease.

UCS is occasionally misdiagnosed as a primum atrial septal defect[3,19]. To differentiate atrial septal defect from a large and dilated CS, it is crucial to note that the former shows a defect in the atrial septum near the atrioventricular valve (AVV), with a cleft in the left AVV. However, a dilated CS, secondary to either a persistent LSVC or anomalous connection of pulmonary veins, appears as a dilation in the posterior part of the right atrium, beyond the level of the AVV. Still, it should not have direct communication with the left atrium unless it is unroofed.

While transthoracic and contrast echocardiography are typically effective in the accurate diagnosis of UCS in most instances[19-24], the condition remains undetected preoperatively in approximately one-third of cases[19]. In our case the absence of the RSVC, the presence of LSVC and its draining site, the roof of the left atrium, the mitral valve pathology and degree of regurgitation as well as the unroofing of the CS were correctly identified by transthoracic and contrast echocardiography. We elected to perform a CT scan to further delineate the exact drainage of the LSVC in relationship to the left upper pulmonary vein and the base of the atrial appendage.

Although surgical repair has traditionally been the standard treatment, innovative percutaneous therapies are progressively emerging as alternatives[25,26]. In our case, the rarity of the condition, the lack of large series, and the absence of a RSVC limited the surgical options; as extra cardiac repair, be it ligation of the LSVC, in the case where a

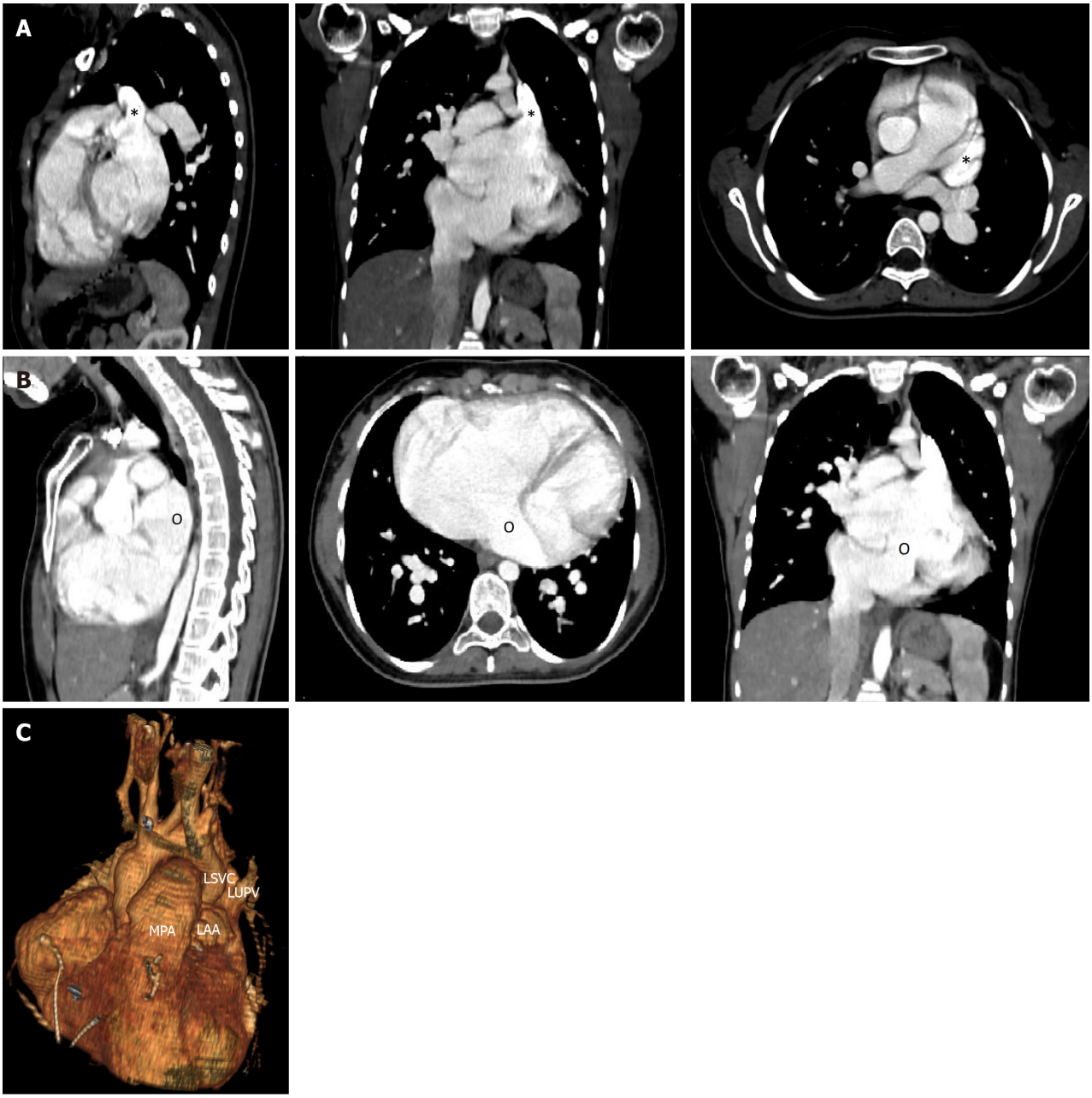

Figure 4 Preoperative computed tomography angiogram. A: The insertion site of the left superior vena cava (LSVC) is shown by an asterisk in the axial, sagittal and coronal views; B: The ostium of the coronary sinus indicated by the letter (o) is shown in the same three views; C: A three-dimensional reconstruction of the LSCV as it enters the left atrium. LSVC: Left superior vena cava; MPA: Main pulmonary artery; LAA: Left atrial appendage; LUPV: Left upper pulmonary vein.

bridging vein is present, or rerouting using a conduit to the RSVC would not be feasible. Mitral valve prolapse and redundant leaflets resulting in severe mitral regurgitation associated with LSVC to UCS may present an additional challenging surgical scenario, as roofing the CS is a concern due to its proximity to the posterior mitral valve leaflet. This proximity could compromise the surgical repair or result in replacement of the mitral valve. In cases of concurrent mitral valve repair, safely diverting the LSVC to the right atrium without obstructing the mitral valve or the pulmonary veins and closure of the CS orifice becomes a crucial aspect of the surgical approach.

Preserving the CS drainage on the left side in the context of mitral valve surgery can offer certain advantages. This simplification potential can reduce overall complexity and operative time while also minimizing the risk of potential complications or obstructions that may arise during redirection. In cases involving mitral valve repair or replacement, opting to avoid roofing of the CS may prove to be a better choice; hence, intra-cardiac baffling without roofing the CS was deemed the procedure of choice in our patient. Post-operative imaging revealed a patent intra-atrial baffle without any significant residual shunt. However, long-term follow-up *via* echocardiography is definitely needed.

Table 1 Published studies describing absent right superior vena cava, unroofed coronary sinus and associated heart lesions, in the absence of heterotaxia syndrome

Case number	Ref.	Associated cardiac disease
1	Brown *et al*[27]	None
2	Wood[28]	None
3	Sherafat *et al*[29]	None
4	Kabbani *et al*[30]	VSD
5	Choi *et al*[31]	DORV
6	Choi *et al*[31]	None
7	Pugliese *et al*[32]	TOF
8	Doksöz *et al*[11]	Cor triatriatum, AVSD
9	Kumar *et al*[12]	Common atrium, AVSD
10	Yilmaz *et al*[13]	Right sided pericardial defect
11	Bitar *et al* (current case)	Myxomatous MV

VSD: Ventricular septal defect; DORV: Double outlet right ventricle; TOF: Tetralogy of Fallot; AVSD: Atrioventricular septal defect; MV: Mitral valve.

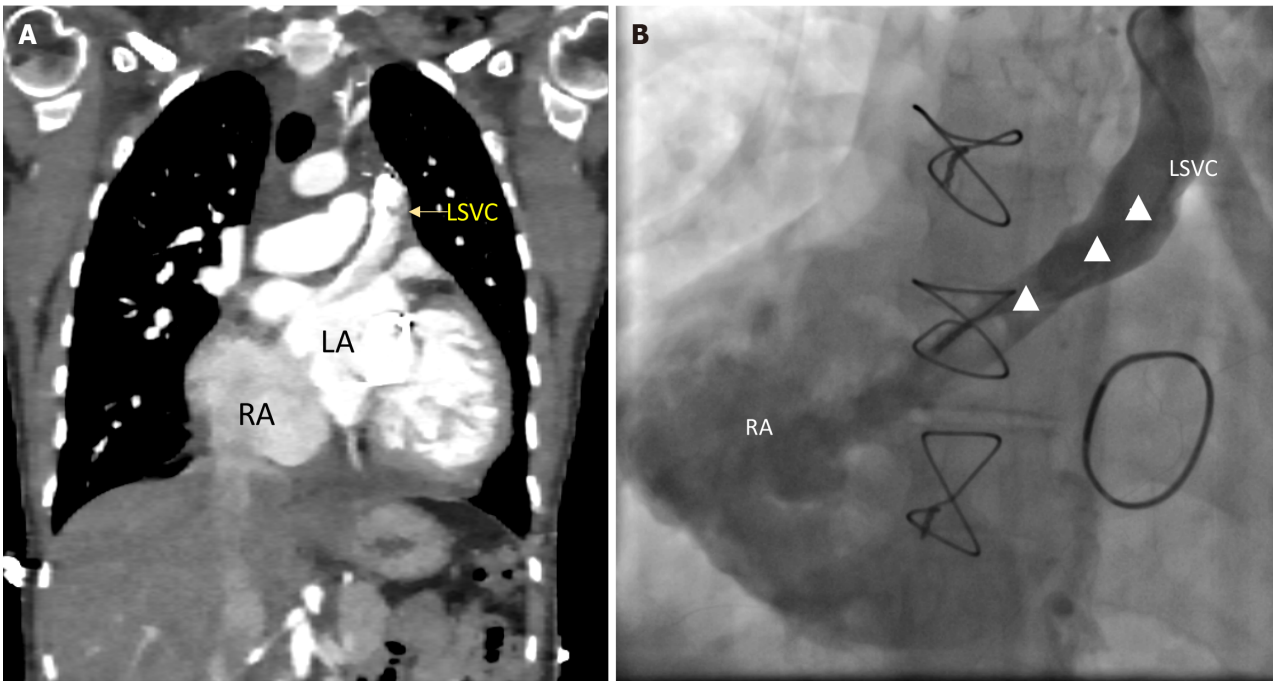

Figure 5 Post-operative imaging. A: Post-operative computed tomography angiogram; B: Antero-posterior venogram of the left superior vena cava (LSVC). Both images show the course of the intra-atrial tunnel (triangle) connecting the LSVC to the right atrium. RA: Right atrium. LA: Left atrium. LSVC: Left superior vena cava.

CONCLUSION

In conclusion, UCS is a complex condition which often requires careful consideration of associated anomalies and tailored surgical approaches, especially in cases involving severe mitral insufficiency, and single LSVC.

FOOTNOTES

Author contributions: Bulbul Z conceived the presented idea and the study framework; Bitar F, Jassar Y, Zareef R, Bulbul Z and Abboud J collected the information and wrote the first draft of the manuscript; Bitar FF, Arabi M and Bulbul Z supervised the project and performed the final editing; All authors contributed to corrections and adjustment of subsequent iterations of the manuscript; All

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ORCID number: Fouad Bitar 0000-0002-4848-4679; Ziad Bulbul 0000-0002-0893-0435; Rana Zareef 0009-0005-3938-8043; Mariam Arabi 0000-0001-6895-1580; Fadi Fouad Bitar 0000-0002-3664-5682.

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