Katetrska ablacija atrijske fibrilacije pri bolnikih s cor triatriatum sinister: opis primera in pregled literature

Anatomical Considerations Regarding Catheter Ablation of Atrial Fibrillation in Cor Triatriatum Sinister: A Case Report and Review of the Literature

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Ključne besede:
cor triatriatum sinister, atrijska fibrilacija, katetrska ablacija

Key words:
cor triatriatum sinister, catheter ablation, atrial fibrillation

Članek prispel / Received
6. 12. 2025
Članek sprejet / Accepted
30. 3. 2026

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Izvleček

Uvod: *Cor triatriatum sinister (CTS) spada med redke prirojene srčne napake. Katetrska ablacija atrijske fibrilacije (AF) pri pacientih s CTS zahteva ustrezno predoperativno pripravo zaradi anatomskih posebnosti.*

Predstavitev primera: *49-letna bolnica z znanim CTS je bila sprejeta v našo ustanovo za katetrsko ablacijo paroksizmalne AF. Ultrazvok srca je prikazal razširjen levi preddvor, ki je bil s tanko membrano razdeljen na dve ločeni votlini. Magnetnoresonančno slikanje srca je razkrilo dodatno membrano pred levimi pljučnimi venami. Poseg smo opravili pod nadzorom znotrajsrčnega ultrazvoka in fluoroskopije. Izdelali smo elektro-anatomsko mapo levega preddvora in električno izolirali desne pljučne vene. Levih pljučnih ven nismo uspeli*

Abstract

Introduction: *Cor triatriatum sinister (CTS) is a rare congenital cardiac defect. Catheter ablation of atrial fibrillation (AF) in patients with CTS has important anatomical considerations.*

Case presentation: *A 49-year old female patient with a known CTS was referred to our institution for catheter ablation of symptomatic paroxysmal AF. Echocardiography revealed a dilated left atrium, which was split into two separate compartments by a thin fibromuscular membrane. A second membranous structure, in front of the left pulmonary vein (PV) ostia, was noted on the preprocedural magnetic resonance scan. The procedure was guided by intracardiac echocardiography and fluoroscopy. A 3D-electroanatomic map was created and the*

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mapirati in izolirati. V več kot enem letu spremljanja po posegu, bolnica simptomov motnje ritma ni več zaznala.

Diskusija: *Katetrska ablacija AF predstavlja uporabno metodo za dolgotrajno vzdrževanje sinusnega ritma pri bolnikih s CTS. V pregledani literaturi so se avtorji pri tej populaciji najpogosteje osredotočili na slikovno diagnostiko ali pred ali med posegom, ter tudi na različne strategije katetrske ablacije. Dodatna membrana pred levimi pljučnimi venami zaenkrat še ni bila opisana.*

right PVs were isolated. The left PVs could not be mapped and isolated. The patient was discharged the following day without complications, and has remained entirely asymptomatic for over one year.

Discussion: *Catheter ablation in patients with AF and a CTS appears to be a feasible rhythm control treatment modality. Most published literature details preprocedural and intraprocedural CTS assessment with different imaging modalities, as well as different ablation strategies based on the arrhythmia type. To the best of our knowledge, an additional membranous structure in front of the left PVs has not been reported.*

INTRODUCTION

Cor triatriatum sinister (CTS) is a rare cardiac anomaly that accounts for approximately 0.1% - 0.4% of congenital cardiac diseases (1). The left atrium is divided into two compartments by a thin fibromuscular membrane. The incidence of atrial fibrillation (AF) in CTS is estimated at around 30% (2). Here, we present a patient with a CTS who underwent catheter ablation for symptomatic paroxysmal AF, and provide a review of the literature regarding catheter ablation in patients with a CTS.

CASE PRESENTATION

A 49-year old female Caucasian patient was referred to our electrophysiology section for catheter ablation of symptomatic paroxysmal AF. At the time of admission, the patient reported episodes of self-limited dyspnea, fatigue and palpitations, lasting for several hours. An electrocardiogram revealed normal sinus rhythm. Transthoracic echocardiography (TTE) showed a mildly dilated left ventricle, normal ejection fraction, and no significant valvular pathologies. The left atrium (LA) was dilated (LAVI = 67 ml/m2) and split into two separate compartments by a thin membrane. Transesophageal echocardiography (TEE) revealed a thin fenestrated fibromuscular membrane that extended from the fossa ovalis to the Coumadin

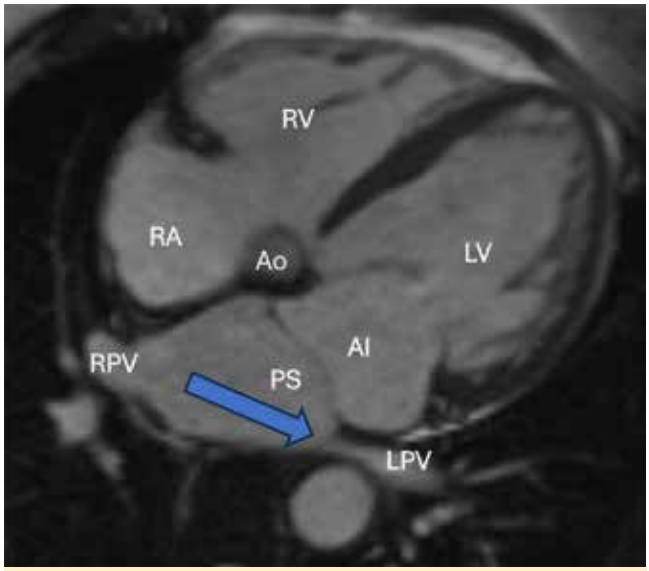

Figure 1: *A 4-chamber view of the cardiac magnetic resonance scan, which shows both compartments of the left atrium. The arrow points towards the second membranous structure, in front of the left pulmonary vein ostium.*

RV – right ventricle, RA – right atrium, LV – left ventricle, Ao – ascending aorta, AI – anteroinferior compartment of the left atrium, PS – posterosuperior compartment of the left atrium, LPV – left pulmonary vein, RPV – right pulmonary vein

ridge. It divided the LA into a posterosuperior section containing pulmonary vein (PV) ostia, and an anteroinferior compartment with the left atrial appendage and the mitral annulus. No significant gradient was noted at the level of the membrane. Cardiac magnetic resonance imaging (MRI) confirmed the position and benign nature of the membrane. Furthermore, a second thin membrane, in front of the left PV ostia, was noted (Figure 1). The left PVs were not dilated and there were no signs of pulmonary congestion.

The procedure was performed in the electrophysiology laboratory under conscious sedation. The patient signed informed consent. Three right femoral vein punctures were performed. A decapolar mapping catheter was placed in the coronary sinus and an intracardiac echocardiography (ICE) probe was placed in the high right atrium. Transseptal puncture (TSP) was guided by fluoroscopy and ICE, and the long sheath was placed in the posterosuperior compartment. A focal contact-force sensing irrigated ablation catheter was inserted and electroanatomic (EAM) mapping was performed (Figure 2). Right PVs were tagged. Left PVs could not be accessed and mapped from the posterosuperior compartment, due to the second membrane. Blood flow from the left PVs towards the posterosuperior compartment of the LA was confirmed with color Doppler on ICE.

Wide antral circumferential point-by-point radiofrequency ablation of the right PVs was performed and was guided by ablation-index (450 on the anterior wall and roof, and 300-350 on the posterior and inferior walls). Bidirectional block to the right PVs was verified. No residual electrograms inside the right PV ostia were observed (entrance block). Exit block was confirmed by a lack of LA capture during high-output pacing maneuvers performed from the inside of the right PVs. A second TSP to the anteroinferior part of the LA could not be performed because the fossa ovalis was oriented entirely towards the posterosuperior chamber. The patient was discharged the following day without complications on antiarrhythmic pharmacotherapy consisting of a low-dose class Ic agent in combination with a beta-adrenergic blocker. A routine outpatient follow-up was scheduled at 12 months.

At follow-up, the ECG demonstrated sinus rhythm and the patient reported no AF-related symptoms.

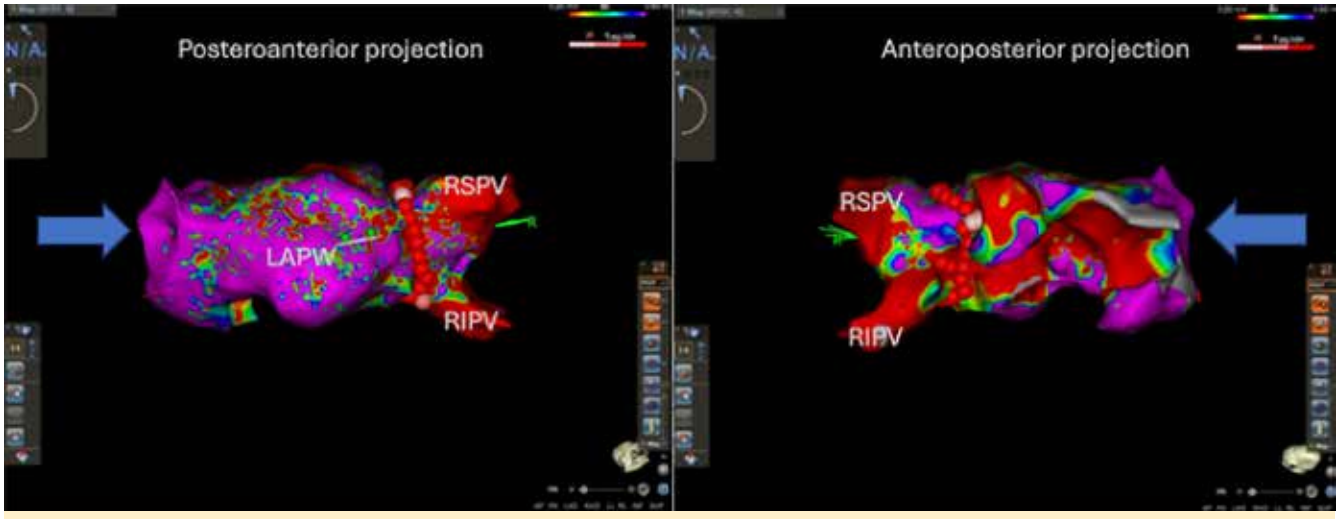

Figure 2: *A 3D-electroanatomic map (posteroanterior projection on the left side and anteroposterior projection on the right side) of the posterosuperior compartment in the left atrium. The arrow points towards a second membranous structure, which prevented mapping of the left pulmonary veins.*

RSPV – right superior pulmonary vein, RIPV – right inferior pulmonary vein, LAPW – left atrial posterior wall

Accordingly, antiarrhythmic therapy was discontinued. Anticoagulation with a direct oral anticoagulant (DOAC) was resumed indefinitely given a CHA2DS2-VA score of 2.

DISCUSSION

Catheter ablation for AF in a patient with a CTS was first described in 2008 (3). Fifteen case reports and two case series (Table 1) have been published to date (2-18). Several reports focused on detailed preoperative imaging techniques (6,9,11,13,17,18) and the utility of ICE or TEE-guided TSP (4,5,7,12,13,15,18). Anatomical considerations for catheter ablation of atrial tachyarrhythmias in patients with CTS were discussed in some studies (4,7,14,15). Coexistence of CTS and other congenital cardiac variations was reported (7,14,15,16). Furthermore, different ablation strategies and modalities were explored in several studies (2,5,8,10,16,17). In summary, catheter ablation has been reported as a feasible rhythm-control strategy in patients with CTS and AF. Nevertheless, meticulous preprocedural anatomic characterization using TTE, TEE, and cardiac CT or MRI is crucial given the altered atrial anatomy associated with CTS. Our case of catheter ablation in a patient with a CTS was similar to previous reports in certain aspects. The LA was split into the posterosuperior section

Table 1: *Procedural characteristics of catheter ablation for left atrial tachyarrhythmias in patients with cor triatritum sinister in our report and in the existing literature.*

Case report/ Case series	Indication	Preprocedural imaging	Guidance of ablation	Transseptal puncture	Ablation targets	Additional information
Yamada et al, 2008 (3)	Paroxysmal AF	TTE, TEE,	Fluoroscopy, 3D EAM	TSP into anteroinferior chamber; ablation catheter looped back through CTS membrane into posterosuperior chamber	PV isolation	First description of AF ablation in CTS; highlighted the need for precise imaging to perform posterior TSP into PV-containing compartment.
Bhatia et al, 2010 (4)	Persistent AF	TTE, CT scan	Fluoroscopy, ICE, 3D EAM (combined with ICE images)	TSP into posterosuperior chamber (ICE-guided)	PV isolation	Feasibility of ICE-guided TSP.
Gavin et al, 2011 (5)	Persistent AF	TTE, CT scan	Fluoroscopy, TEE, 3D EAM	TSP into posterosuperior chamber (TEE-guided)	PV isolation, roof line, mitral isthmus line, complex fractionated electrogram ablation	Feasibility of TEE-guided TSP; complex ablation procedure in both compartments of LA.
Fukumoto et al, 2012 (6)	Paroxysmal AF	TTE, TEE, CT scan	Fluoroscopy, ICE, 3D EAM (merged with CT scan)	TSP into posterosuperior chamber (ICE-guided)	PV isolation	Merging 3D EAM with CT scan in CTS.
Tokuda et al, 2016 (7)	Persistent AF	TTE, CT scan	Fluoroscopy, ICE, 3D EAM	No TSP, access to posterosuperior compartment obtained via patent foramen ovale	Left common trunk and right PVs isolation, box lesion in posterior LA	Coexistent CTS and left common PV trunk.

Case report/ Case series	Indication	Preprocedural imaging	Guidance of ablation	Transseptal puncture	Ablation targets	Additional information
Borne et al, 2016 (8)	AF and macroreentrant AT (perimitral, roof-dependent, CTI-dependent flutter)	TTE, TEE, CT scan	Fluoroscopy, ICE, 3D EAM	TSP into posterosuperior chamber (ICE-guided)	PV isolation, roof line, mitral isthmus line, CTI line	Sequential treatment of AF and multiple macroreentrant ATs in CTS.
Paelinck et al, 2018 (9)	Paroxysmal AF	TTE, CT scan	Fluoroscopy, TEE, 3D EAM	TSP into posterosuperior chamber (TEE-guided)	PV isolation	/
Shah et al, 2018 (10)	Long-standing persistent AF	TTE, TEE, CT scan	ICE and 3D EAM	TSP into posterosuperior compartment (ICE-guided)	PV isolation	Zero-fluoroscopy ablation in CTS.
Morishima et al, 2020 (11)	Paroxysmal AF	TTE, CT scan	Fluoroscopy, ICE and 3D EAM (merged with ICE images)	TSP into posterosuperior chamber (ICE-guided)	PV isolation, SVC isolation	/
Okada et al, 2020 (12)	Macroreentrant atrial tachycardia (incorporating fibromuscular membrane)	TTE, CT scan	Fluoroscopy,ICE and 3D EAM	TSP into posterosuperior chamber (ICE-guided)	PV isolation, SVC isolation, ablation of fractionated potentials on the membrane	Figure-of-8 re-entry using CTS membrane as a critical isthmus.
Lugtu et al, 2021; case series (13)	AF or macroreentrant AT (4 patients; case series)	TTE, CT scan	Fluoroscopy, 3D EAM (merged with cardiac CT)	TSP into posterosuperior chamber (TEE or fluoroscopy-guided)	PV isolation (4 patients), roof-line and a line from the membrane to the roof-line (1 patient)	Fractionated potentials on the fibromuscular membrane.
Karimianpour et al; case series 2021 (2)	AF (four patients; case series), macroreentrant atrial tachycardia (one patient)	TTE, TEE (2 cases), CT scan	Fluoroscopy, ICE, 3D EAM	TSP into posterosuperior chamber (ICE-guided)	PV isolation (4 patients), roof-line, mitral isthmus line, ablation of fractionated potentials next to the membrane (1 patient)	CTS present in 0.1% of AF ablation refferals; reisolation of PVs in recurrent AF is feasible.
Minciuna et al, 2022 (14)	Persistent AF	TTE, CT scan	Fluoroscopy, TEE, 3D EAM	TSP into posterosuperior chamber (TEE-guided)	Left common trunk and right PVs isolation; high-power short-duration RF energy.	Catheter stability issues when ablating on common trunk near the CTS membrane.
Iwata et al, 2022 (15)	Persistent AF	TTE, TEE, CT scan	Fluoroscopy, TEE, 3D EAM	TSP into posterosuperior chamber (TEE-guided)	PV isolation and SVC isolation	Coexistent CTS and ASD, feasibility of AF ablation in an octogenerian patient.

Case report/ Case series	Indication	Preprocedural imaging	Guidance of ablation	Transseptal puncture	Ablation targets	Additional information
Kolakowski et al, 2023 (16)	Paroxysmal AF	TTE, CT scan	Fluoroscopy, TEE,	TSP into posterosuperior chamber (TEE-guided)	Single-shot cryoballoon PV isolation	Cryoablation in CTS, concomitant bicuspid aortic valve and CTS.
Lai et al, 2024 (17)	Persistent AF	TTE, TEE, CT scan	Fluoroscopy, ICE, 3D EAM	TSP into posterosuperior chamber (ICE-guided)	PV isolation, LA box lesion set, mitral isthmus line, vein of Marshall ethanol infusion, CTI line	Vein of Marshall ethanol infusion in patients with CTS.
Okuyama et al, 2024 (18)	Persistent AF	TTE, TEE, CT scan	Fluoroscopy, ICE, 3D EAM	TSP into posterosuperior chamber (ICE-guided)	PV isolation, SVC isolation, CTI line	/
Present case	Paroxysmal AF	TTE, TEE, MRI scan	Fluoroscopy, ICE, 3D EAM	TSP into posterosuperior chamber (ICE-guided)	Isolation of right PVs; isolation of left PVs could not be achieved	Additional separate membrane in front of left PV ostia verified by cardiac MRI and ICE.

AF – atrial fibrillation, TTE – transthoracic echocardiogram, TEE – transesophageal echocardiogram, TSP – transseptal puncture, EAM – electroanatomic mapping, CTS – cor triatritum sinister, PV – pulmonary vein, CT – computed tomography, ICE – intracardiac echocardiography, LA – left atrium, CTI – cavotricuspid isthmus, SVC – superior vena cava, AT – atrial tachycardia, MRI – magnetic resonance imaging

with PV ostia, and the anteroinferior section with mitral annulus and left atrial appendage. Contrary to the previous reports, the left PVs could not be mapped from the posterosuperior compartment of the LA, despite the use of a focal mapping and ablation catheter enabling point-by-point mapping to optimize maneuverability. Preprocedural MRI scan and ICE both revealed a separate thin membrane in front of the left PV ostia, which prevented catheter manipulation and access to the left PVs. Color Doppler on ICE revealed blood flow from the left PVs to the posterosuperior compartment, the left PVs were not dilated on the preprocedural MRI scan and there were no clinical signs of pulmonary congestion. Therefore, the blood flow into the posterosuperior compartment of the LA was not significantly obstructed. To the best of our knowledge, such a structure has not been reported previously in a case of catheter ablation in a patient with a CTS. Although only isolation of the right PVs could be performed, the patient has remained entirely asymptomatic for over 13 months. As the EAM of the left PVs could not be performed, their electrical activity could not be characterized. Consequently, the pathophysiological basis for the observed procedural success remains uncertain. Nevertheless, asymptomatic recurrence of AF cannot be excluded.

CONCLUSIONTS

Catheter ablation in patients with CTS and AF is a feasible and safe rhythm control treatment modality. However, comprehensive preprocedural anatomic characterization using complementary imaging modalities is essential given the altered atrial anatomy associated with CTS. An additional membranous structure, in front of the left PV ostia, may impede catheter access to the left PVs.

GRANT SUPPORT

No funding was received to assist with the preparation of this manuscript.

CONFLICT OF INTEREST DISCLOSURE

Authors have nothing to disclose.

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