ESC European Society of Cardiology

European Heart Journal - Case Reports (2023) **7**, 1–4
https://doi.org/10.1093/ehjcr/ytac481

CASE REPORT
Electrophysiology

A case report of paroxysmal atrial fibrillation in three pulmonary veins presenting a common trunk

Hirofumi Kujiraoka *, Rintaro Hojo, Masao Takahashi, and Seiji Fukamizu

Department of Cardiology, Tokyo Metropolitan Hiroo Hospital, 2-34-10, Ebisu, Shibuya-ku, Tokyo 150-0013, Japan

Received 12 July 2022; first decision 11 August 2022; accepted 14 December 2022; online publish-ahead-of-print 17 December 2022

Background The pulmonary vein (PV) variant is present in 23–38% of patients who undergo atrial fibrillation ablation, and the common inferior PV (CIPV) variant is a rare PV variant that has been reported in 0.9–1.5% of patients. The arrhythmogenicity of the common trunk of the CIPV is unknown.

Case summary A 77-year-old woman underwent catheter ablation for paroxysmal atrial fibrillation (AF). Preoperative computed tomography revealed a common trunk from which the bilateral inferior PVs and a left superior PV originated. The voltage map of the left atrium (LA) showed three PVs stemming from a common trunk. There was a low-voltage area bounded by the common trunk entrance. An isolation line was created to connect the right superior PV and the common trunk. Twelve months later, AF recurred. The voltage map in second session showed residual irregular potentials at the boundary between the common trunk and the LA, and posterior wall isolation was performed. Postoperatively, the patient maintained sinus rhythm with no antiarrhythmic drugs during the 12-month follow-up period.

Discussion The CIPV is likely to predict the AF recurrence, even if preoperative voltage mapping shows a low voltage area. Substrate modification should be performed on abnormal potentials at the entrance of the common trunk, even though no potential is detected in the PVs or their antrum.

Keywords Paroxysmal atrial fibrillation • Case report • Computed tomography • Catheter ablation

ESC Curriculum 2.4 Cardiac computed tomography • 5.3 Atrial fibrillation

Learning points

- A case in which the left superior pulmonary vein (PV) and both inferior PVs present a common trunk is a rare anomaly.
- The common inferior PV variant is likely to predict the AF recurrence, even if preoperative voltage mapping shows a low voltage area.
- In three common PV case, substrate modification should be performed on abnormal potentials at the entrance of the common trunk, even though no potential is detected in the PVs or their antrum.

Introduction

Patients referred for atrial fibrillation (AF) ablation often have anatomical variants that can influence the procedure. The pulmonary vein (PV) variant is present in 23–38% of patients who undergo AF ablation, and the common inferior PV (CIPV) variant is a rare PV variant that has been reported in 0.9–1.5% of patients.[1,2] It has been reported that PV isolation (PVI) for CIPV is difficult in terms of morphological

* Corresponding author. Tel: +81-3-3444-1181, Fax: +81-3-3444-3196, Email: kujira1623@gmail.com
Handling Editor: Patrick Badertscher
Peer-reviewers: Marta Cvijic; Richard Ang
Compliance Editor: Ralph Mark Louis Neijenhuis
Supplementary Material Editor: Jonathan Senior

features.[3] We experienced a paroxysmal AF case in which three PVs presented a common trunk and reported on the necessary points for catheter ablation.

Timeline

Time	Events
2018, May	Patient developed atrial fibrillation (AF)
2018, August	Patient underwent pulmonary vein isolation(PVI)
2019, August	Patient had AF recurrence
2019, September	Patient underwent second session for AF recurrence. Posterior wall isolation was performed. No PV reconduction was recorded.
2020, September	Patient was followed up with 12-lead ECG and Holter ECG, and no arrhythmia was reported.

Case report

The patient was a 77-year-old woman with a history of hypertension and dyslipidaemia. She was taking amlodipine and rosuvastatin. She presented to her local doctor with symptoms of presyncope. Holter electrocardiography (ECG) showed paroxysmal AF and sinus arrest after the termination of tachycardia. Consequently, the patient underwent catheter ablation for paroxysmal AF. The study participant provided informed consent. Pre-operative contrast-enhanced computed tomography (CT) was performed to evaluate for thrombus in the left atrium (LA) and to assess the morphology of the LA. It revealed a common trunk from which the bilateral inferior PVs and a left superior PV originated (*Figure 1A and B*).

We created a voltage map of the LA during pacing from the right atrium using a three-dimensional (3D) mapping system (CARTO-3 system version 6; Biosense Webster Inc., Diamond Bar, CA, USA) and a 20-electrode mapping catheter (PENTARAY; Biosense Webster, Diamond Bar, California) using a trans-septal approach. A voltage range of less than 0.5 mV was defined as a low voltage area.[4] The three PVs stemming from a common trunk showed a low-voltage area bounded by the common trunk entrance (*Figure 1C*), which was not captured by electrical stimulation. As AF was noted to occur due to firing from the right superior PV (RSPV) (*Figure 2A*), we created an isolation line to connect the RSPV and the common trunk (*Figure 2B and C*) with point-by-point RF applications at 40 W using the contact force–sensing irrigated ablation catheter (ThermoCool SmartTouch SF, Biosense Webster). The session was terminated after confirming that the potentials of the four PVs had disappeared. The patient was followed up with 12-lead ECG and Holter ECG.

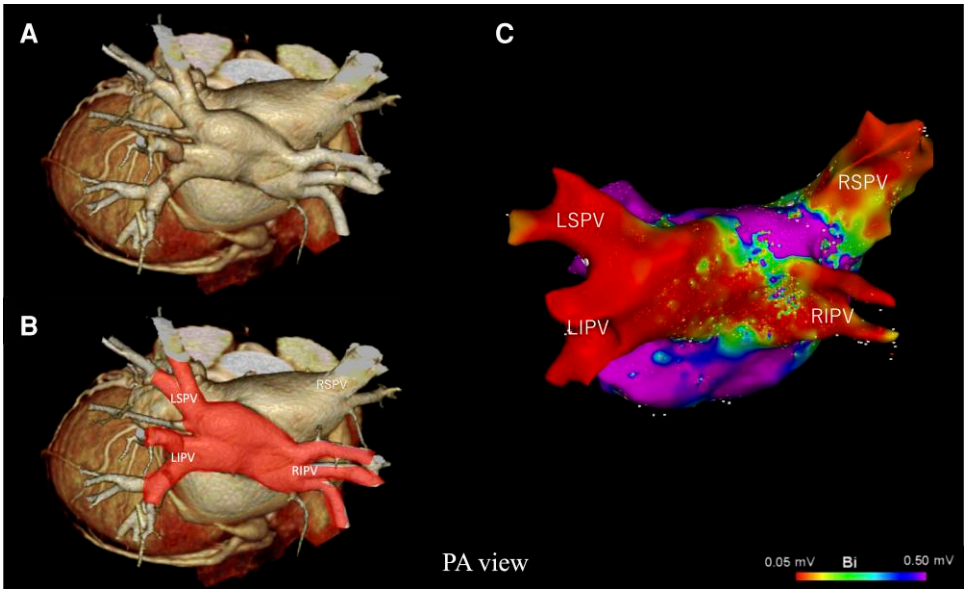

Figure 1 (*A* and *B*) Pre-procedural cardiac computed tomography demonstrating right inferior pulmonary vein, left inferior pulmonary vein, and left superior pulmonary vein have one common trunk. (*C*) Left atrium voltage map before ablation obtained during pacing from the right atrium shows that the common pulmonary veins were low voltage area bounded by the common trunk entrance.

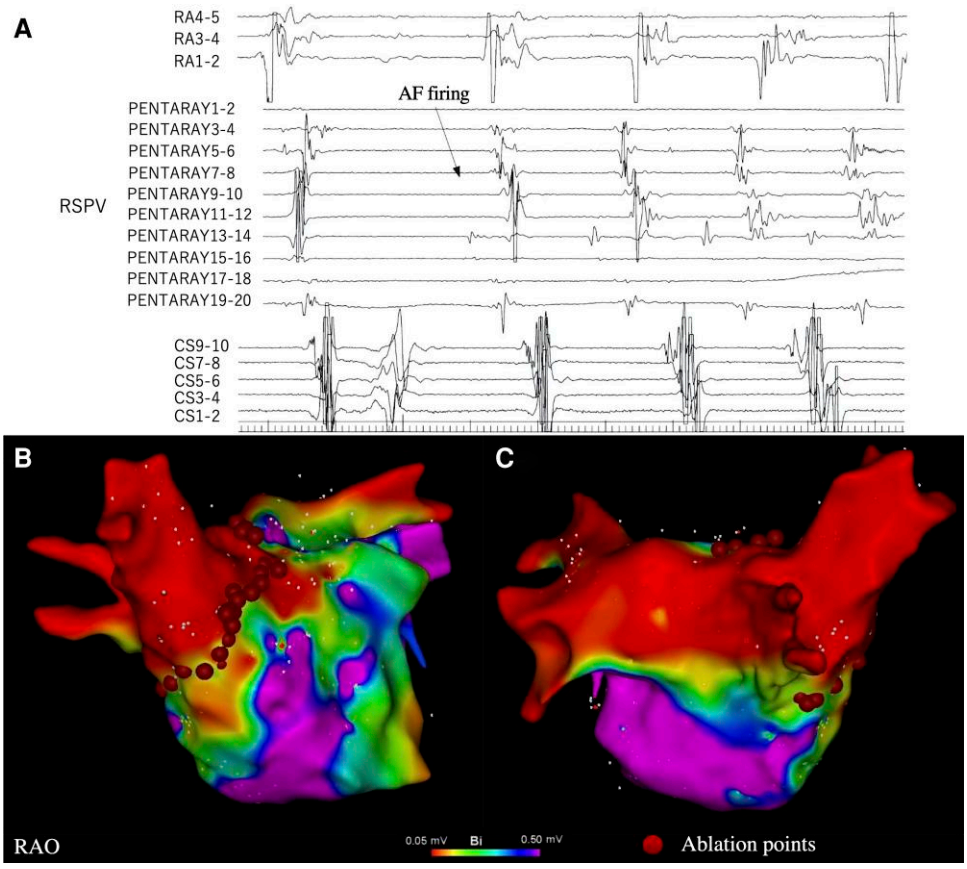

Figure 2 (*A*) Atrial fibrillation occurring upon ectopic firing from right superior pulmonary vein. (*B* and *C*) Left atrium voltage map post-ablation in the first session. An isolation line was created to connect the right superior pulmonary vein and the common trunk. The common trunk and four pulmonary veins show low voltage areas. AP, anteroposterior; CS, coronary sinus; PA, posteroanterior; RA, right atrium.

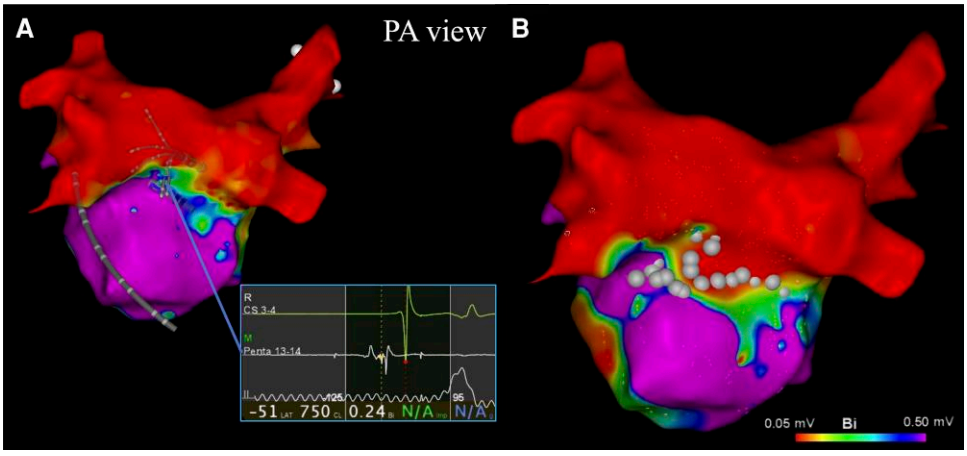

Figure 3 (*A*) The left atrium voltage map before ablation in the second session. Residual irregular potential remains at the border between the common trunk and left atrium. (*B*) The left atrium voltage map post-ablation. Substrate modification was accomplished by creating a bottom line. PA, posteroanterior.

Twelve months later, paroxysmal AF recurred, and a second catheter ablation session was performed. Although no LA-PV reconduction was detected, the voltage map showed residual irregular potentials at the boundary between the common trunk and the LA (*Figure 3A*). Consequently, we isolated the posterior wall of the LA, including the boundary between the LA and common trunk. Posterior wall isolation (*Figure 3B*) was conducted using a pentaray catheter after high-dose isoproterenol provocation and confirmed through voltage mapping and pacing with an exit block. Post-operatively, the patient maintained sinus rhythm with no antiarrhythmic drugs during the 12-month follow-up period.

Discussion

Embryologically, the common PV joins the primary atrial component of the heart and begins to bifurcate, dilate, and incorporate into the LA at a gestational age of approximately 30 days. By 7–8 weeks of gestation, contemporaneously with septation of the atria and ventricles, the common PV separates into the right PV and left PV.[5] During this developmental process, an incomplete PV incorporation can result in a unilateral common PV ostium, whereas over-incorporation can lead to more than four PV ostia.[6] Considering the mechanism of PV development, a case in which the left superior PV and both inferior PVs present a common trunk is a rare anomaly and has never been reported with detailed 3D mapping.

The PVs contain myocardial sleeves extending from the LA,[7] which consist of a combination of working cardiomyocytes and pacemaker cells.[8] Ectopic electrical activity originating from PV myocardial sleeves has been found to trigger and maintain paroxysmal AF in humans.[9] Although the common left PV is known to be a consistent source of arrhythmogenic atrial ectopy, the arrhythmogenicity of the CIPV has not been evaluated systematically. In a review of 11 paroxysmal AF cases presenting with CIPV, a local AF trigger was observed in 57% of the CIPV cases, and an effective strategy was circumferential PV electrophysiological isolation with 'tri-circle': one surrounds the LSPV ostium, one surrounds the RSPV ostium, and the other surrounds the common ostium of two inferior PVs.[10] Another similar study, in which an AF trigger potential was recorded within the CIPV, demonstrated the success of a tri-circle PVI.[11] Conversely, Yamane *et al.*[12] reported that CIPV rarely originates from the extrasystole that triggers AF. Moreover, it has been reported that CIPV presents as an extensive low-voltage area. Thus, it is controversial whether empirical electrical isolation of the CIPV should be performed.

In the present case, the common trunk displayed a low voltage area in the voltage map before PVI. There was no conduction between the common trunk and LA. PVI was achieved by a block line connecting the RSPV and the common trunk. However, AF recurred during the chronic phase. In the second session, substrate modification was performed for a residual irregular potential, which was observed in the border region between the common trunk and posterior wall.

The findings from this case demonstrate that CIPV is likely to predict the AF recurrence, even if preoperative voltage mapping shows a low voltage area. Although the common trunk has a low voltage area, ectopic electrical activity may occur from the area within the PV myocardial sleeve at the LA border. It is reported that non-PV triggers of AF are likely to arise from in or around low-voltage areas in the LA.[13] Substrate modification should be performed on abnormal potentials at the entrance of the common trunk, even though no potential is detected in the PVs or their antrum.

Conclusion

We experienced a paroxysmal AF case in which three PVs presented a common trunk.

Lead author biography

Hirofumi Kujiraoka was born in Ibaraki, Japan, in 1991. He received MD degree from University of Tsukuba School of Medicine, Ibaraki, Japan, in 2016, and completed 2 years of Japanese general residency training at National Tokyo Medical Center. Since 2021, Dr Kujiraoka has been working as a cardiologist at Tokyo Metropolitan Hiroo Hospital.

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: Written informed consent in accordance with COPE guidelines was obtained from the patient.

Conflict of interest: None declared.

Funding: None declared.

References

1. Kanaji Y, Miyazaki S, Iwasawa J, Ichihara N, Takagi T, Kuroi A, *et al.* Pre-procedural evaluation of the left atrial anatomy in patients referred for catheter ablation of atrial fibrillation. *J Cardiol* 2016;**67**:115–121.
2. Yamane T, Date T, Sugimoto K, Mochizuki SE. Confluent inferior pulmonary veins in a patient with paroxysmal atrial fibrillation. *J Cardiovasc Electrophysiol* 2005;**16**:107.
3. Canpolat U, Yorgun H, Hazirolan T, Aytemir K. Common ostium of inferior pulmonary veins: an extremely rare variant described by preprocedural computerized tomography angiography. *J Arrhythm* 2021;**37**:450–451.
4. Verma A, Wazni OM, Marrouche NF, Martin DO, Kilicaslan F, Minor S, *et al.* Pre-existent left atrial scarring in patients undergoing pulmonary vein antrum isolation: an independent predictor of procedural failure. *J Am Coll Cardiol* 2005;**45**:285–292.
5. Webb S, Kanani M, Anderson RH, Richardson MK, Brown NA. Development of the human pulmonary vein and its incorporation in the morphologically left atrium. *Cardiol Young* 2001;**11**:632–642.
6. Douglas YL, Jongbloed MR, Deruiter MC, Gittenberger-de Groot AC. Normal and abnormal development of pulmonary veins: state of the art and correlation with clinical entities. *Int J Cardiol* 2011;**147**:13–24.
7. Nathan H, Eliakim M. The junction between the left atrium and the pulmonary veins. An anatomic study of human hearts. *Circulation* 1966;**34**:412–422.
8. Cheung DW. Electrical activity of the pulmonary vein and its interaction with the right atrium in the Guinea-pig. *J Physiol* 1981;**314**:445–456.
9. Haissaguerre M, Jais P, Shah DC, Takahashi A, Hocini M, Quiniou G, *et al.* Spontaneous initiation of atrial fibrillation by ectopic beats originating in the pulmonary veins. *N Engl J Med* 1998;**339**:659–666.
10. Yu R, Dong J, Zhang Z, Liu X, Kang J, Long D, *et al.* Characteristics in image integration system guiding catheter ablation of atrial fibrillation with a common ostium of inferior pulmonary veins. *Pacing Clin Electrophysiol* 2008;**31**:93–98.
11. Itoh N, Maruyama M, Tsuboi I, Nohara T, Ishihara S, Shimizu W, *et al.* Arrhythmogenic triggers of atrial fibrillation arising from the common trunk of the inferior pulmonary veins. *Circ Rep* 2022;**4**:147–148.
12. Yamane T, Date T, Tokuda M, Aramaki Y, Inada K, Matsuo S, *et al.* Prevalence, morphological and electrophysiological characteristics of confluent inferior pulmonary veins in patients with atrial fibrillation. *Circ J* 2008;**72**:1285–1290.
13. Kawai S, Mukai Y, Inoue S, Yakabe D, Nagaoka K, Sakamoto K, *et al.* Non-pulmonary vein triggers of atrial fibrillation are likely to arise from low-voltage areas in the left atrium. *Sci Rep* 2019;**9**:12271.