

A case report of an important role of epicardial connections in producing spontaneous pulmonary vein activity and initiating and maintaining atrial fibrillation

Keishiro Yagyu, MD, Masao Takemoto, MD, PhD, Masaki Fujiwara, MD, PhD, Yoshibumi Antoku, MD, PhD, Tokushi Koga, MD, PhD, Takuya Tsuchihashi, MD, PhD

From the Cardiovascular Center, Steel Memorial Yawata Hospital, Kitakyushu, Japan.

Introduction

Pulmonary vein antrum isolation (PVAI) with radiofrequency catheter ablation (RFCA) has proven to be a useful strategy for atrial fibrillation (AF) worldwide.^{1–3} To prevent initiating and maintaining AF, a complete PVAI should be a target of the treatment of AF.¹ However, even if using contemporary advanced techniques such as a 3dimensional mapping system and contact force monitoring, a first-pass isolation is sometimes difficult to perform and additional ablation inside the veins is commonly necessary and effective in achieving these objectives.⁴ Recent studies have demonstrated that the presence of epicardial connections (ECs)⁵ involving the pulmonary veins (PVs) is one of the mechanisms for failure in achieving a complete PVAI.^{6–8}

Case report

Written consent for the submission and publication of this case report, including the images and associated text, has been obtained from the patient in line with COPE guidance.

A 75-year-old male patient was admitted to our hospital to undergo RFCA of paroxysmal AF. He had a history of hypertension. Anticoagulation, antiarrhythmic, and antihypertensive therapies with apixaban 10 mg, bepridil hydrochloride 100 mg, and telmisartan 40 mg daily had already been started. On admission, his blood pressure and heart rate were 114/72 mm Hg and 86 beats per minute, respectively. Auscultation revealed normal cardiac and breath sounds. His serum creatinine and brain natriuretic peptide levels were 1.04 mg/dL and 190.0 pg/mL, respectively. The 12-

KEYWORDS Atrial fibrillation; Case report; Catheter ablation; Pericardial connection; Spontaneous activities (Heart Rhythm Case Reports 2022;8:643–649)

KEY TEACHING POINTS

- Recent studies have demonstrated that the presence of epicardial connections (ECs) involving pulmonary veins (PVs) are one of the mechanisms for failure in achieving a complete pulmonary vein antrum isolation (PVAI) of atrial fibrillation (AF).
- ECs may possibly play an important role in producing spontaneous activities and initiating and maintaining AF.
- The physicians should be aware of the possibility of the presence of ECs even though the conventional PVAI is completed, when performing ablation of AF.

lead electrocardiogram and chest radiograph exhibited sinus rhythm with complete right bundle branch block (Figure 1A) and a normal cardiothoracic ratio (Figure 1B). The echocardiography yielded a normal left ventricular ejection fraction of 55%, normal valvular function, and no evidence of structural heart disease. His left atrial (LA) dimension was 38.3 mm. His CHADS₂/CHA₂DS₂-VASc score was 3/4, respectively. Transesophageal echocardiography demonstrated the absence of any LA thrombi the day before the RFCA. Before the RFCA, because the AF continued, an intracardiac electrical conversion was performed using 30 J with a deflectable catheter, BeeAT (Japan Lifeline, Tokyo, Japan), placed in the coronary sinus (CS) and right atrium in order to restore sinus rhythm. However, sinus rhythm could not be maintained and AF instantaneously recurred. Then, during AF, following a double transseptal puncture guided by intracardiac echocardiography (ViewFlex Xtra ICE catheter; Abbott, Plymouth, MN), a circumferential PVAI was performed under electroanatomic guidance with a 3D mapping system (EnSite; St. Jude Medical, St. Paul, MN) using a TactiCath SE open irrigated ablation catheter (Abbott, Plymouth, MN) through a steerable introducer (Agilis NxT; Abbott,

Funding Sources: None. Disclosures: All authors have nothing to disclose. Address reprint requests and correspondence: Dr Masao Takemoto, Cardiovascular Center, Steel Memorial Yawata Hospital, 1-1-1 Haruno-machi, Yahatahigashi-ku, Kitakyushu 805-8508, Japan. E-mail address: matakemo@kc4.so-net.ne.jp; matakemo@cardiol.med.kyushu-u. ac.jp.



Figure 1 A 12-lead electrocardiogram on admission (**A**) and during the procedure (**H**), and chest radiograph on admission (**B**). **C**: The fluoroscopic anteriorposterior image during the procedure. **D–G**: The EnSite (St. Jude Medical, St. Paul, MN) image in the posterior-anterior (**D**) and anteroposterior (**E**, **F**, and **G**) views of the patient during the procedure. The white arrows indicate the remaining potential in the LSPV. ABL = ablation catheter; CMC = circular mapping

Plymouth, MN) under the administration of isoproterenol $(10-20 \mu g/h)$. When the circular mapping catheter (CMC) (Optima; Abbott, Plymouth, MN) was placed just inside the PVAI lines, there were no remaining potentials detected with the CMC in all 4 PVs (Figure 2A). However, after the PVAI, sinus rhythm could still not be maintained. Thus, a LA posterior wall isolation was additionally performed. However, sinus rhythm still could not be maintained. Because we suspected the presence of ECs,^{5,6} the LA and PVs were reconstructed in detail by the EnSite system using a steerable high-density mapping catheter (Advisor HD Grid catheter; Abbott, Plymouth, MN) and demonstrated a remaining potential at a 35-mm-distal site from the PVAI line in the left superior pulmonary vein (LSPV; white arrows in Figure 1D and 1E). We also confirmed there were no concealed low-voltage signals (CLVSs)⁹ along the PVAI lines, indicating the completion of the PVAI lines. A CMC also could detect not only this potential but also spontaneous activity¹⁰ in the LSPV (Figure 1F and yellow arrows in Figure 2B). After a spontaneous recovery of sinus rhythm, AF recurred instantaneously, initiated by spontaneous activity¹⁰ in the LSPV (yellow arrow in Figure 2C). We confirmed that the sites of the spontaneous activity and remaining potential were at the same position in the LSPV. Radiofrequency energy was delivered at this site with a maximum power of 25 W (Figure 1C and 1F and yellow arrow in Figure 2D). Then, the AF steadily terminated and sinus rhythm was recovered on the 12-lead electrocardiogram (Figure 1H and red arrows in Figure 2D and 2E), but the AF continued on the intracardiac electrocardiogram of the LSPV (Figure 1H and white arrows in Figure 2D and 2E). A continuous radiofrequency energy delivery at this site completely abolished not only the AF but also the remaining potential and spontaneous activity in the LSPV (blue arrow in Figure 2E and 2F). Then, using the high-density mapping catheter, we confirmed in detail that there were no remaining potentials or spontaneous activity in the 4 PVs or CLVSs⁹ along the PVAI lines. Further, entrance and exit block were also confirmed by LA pacing and pacing just within the PV ostium. Finally, we could achieve bidirectional conduction block between the LA and PVs. During a 30-minute observation period, no spontaneous activity in the LSPV was observed (Figure 2F). Thereafter, programmed stimulation could no longer induce any arrhythmias, including spontaneous activities or AF under the administration of isoproterenol (10-20 µg/h). The patient has remained well, without any arrhythmias or symptoms, for 2 years after the RFCA.

Discussion

After the RFCA of the remaining potential in the LSPV, we confirmed in detail that there were no remaining potentials in

the 4 PVs or CLVSs⁹ along the PVAI lines using the highdensity mapping catheter, and entrance and exit block were confirmed by LA pacing and pacing just within the PV ostium (Figure 2F). Thus, we could definitely achieve a complete conventional PVAI before the RFCA of the remaining potential. Therefore, the remaining potentials in the LSPV were the conduction of the EC, not a gap conduction. Additionally, the LA appendage (LAA) in this present case was not so big, and the distance between the LAA and remaining potentials in the LSPV was 11 mm (Figure 1G). Further, no far-field potentials from the LAA were observed in the LSPV after the RFCA of the remaining potential (EC) (Figure 2F). Thus, the potentials that were recorded in the LSPV before the RFCA of the remaining potential were definitely the potentials of the EC.

On the other hand, before the RFCA of the remaining potential, irregular spontaneous activities were observed at the same position as the remaining potential in the LSPV during AF (Figure 1F and yellow arrows in Figure 2B). If those activities were caused by atrial tachycardias (ATs), the activities of the ATs would have been suppressed during AF. However, the irregular spontaneous activities continued during AF (Figure 2B). These findings were unlikely to be ATs, but were more likely to be spontaneous activities. Moreover, a previous report demonstrated that an initiation of sustained or nonsustained local fibrillation was recorded in cases with a common left-sided PV with preceding automatic activity following the completion of the PVAI.¹¹ Although this present case was not a common left-sided PV, the same phenomenon might have been observed in this present case.

Then, AF recurred instantaneously, initiated by the spontaneous activity in the LSPV (yellow arrow in Figure 2C) after spontaneous recovery of sinus rhythm. Thus, because we suspected the presence of ECs,⁶ the LA and PVs were reconstructed in detail by the EnSite system using the high-density mapping catheter, and it demonstrated a remaining potential 35 mm distal from the PVAI line in the LSPV (Figure 1C-1F). Because the location of the remaining potential was more than 5 mm away from the conventional PVAI line, a gap conduction was unlikely,⁶ indicating it was the conduction of the EC(s). Because the sites of the spontaneous activity and remaining potential were in the same position in the LSPV, the EC might have produced the spontaneous activity contributing to the initiation of AF. The radiofrequency energy delivering for this remaining potential could steadily terminate the AF on the 12-lead electrocardiogram (Figure 1H and white arrows in Figure 2D and 2E) and subsequently on the intracardiac electrocardiogram of the LSPV (blue arrow in Figure 2E). Thus, the EC ran through and conducted the AF from the LSPV to the atrium somewhere and could maintain the AF in both the LSPV and

catheter; CS = coronary sinus; His = His bundle; HRA = high right atrium; LAA = left atrium; LAA = left atrial appendage; LIPV = left inferior pulmonary vein; LSPV = left superior pulmonary vein; MA = mitral annulus; RIPV = right inferior pulmonary vein; RSPV = right superior pulmonary vein; RV = right ventricle.



Figure 2 The intracardiac electrocardiograms after a conventional pulmonary vein antrum isolation (PVAI) (\mathbf{A}), from a circular mapping catheter placed at a site distal to the PVAI line in the left superior pulmonary vein (\mathbf{B}), after spontaneous recovery of sinus rhythm (\mathbf{C}), during the radiofrequency energy delivery (\mathbf{D} and \mathbf{E}), and after completing the PVAI (\mathbf{F}).

atrium. After a radiofrequency energy delivery to the remaining potential at a distal LSPV site, the conduction from the LSPV to the atrium and spontaneous activity in the LSPV were no longer observed (Figure 2E and 2F) during a 30-minute observation period. In view of these findings, the EC in this present case might have played an important role in producing the spontaneous activity and initiating and maintaining the AF.

Although we could not confirm where the EC conducted to from the LSPV in this present case, it might possibly



have conducted to the CS or Marshall bundle because a recent comprehensive study demonstrated that all ECs involving left-sided PVs are associated with the CS or Marshall bundle.⁶ We also could not confirm whether the EC was unidirectional or bidirectional. However, there was certainly at least unidirectional conduction from the PV to somewhere in the atrium. A previous report demonstrated that unidirectional entrance block with spontaneous activity

in the PVs may not be a good indication of a complete PVAI and bidirectional block can reduce the acute PV reconnections and may reduce the AF recurrence in patients undergoing a PVAI of AF.¹⁰ Moreover, the EC in this present case might have played an important role in producing the spontaneous activity and initiating and maintaining the AF. These mechanism(s) of ECs may be a cornerstone in recurrences of AF in patients with ECs undergoing a PVAI of AF.





Finally, because we could achieve bidirectional block between the PV and atrium by RFCA of the remaining potential in the LSPV and confirm the noninducibility of AF, we thought the AF recurrence rate might have been low in this present case. Actually, this patient has remained well, without AF recurrence or symptoms for 2 years after the RFCA.

Conclusion

To the best of our knowledge, this is the first report that ECs may possibly play an important role in producing spontaneous activity and initiating and maintaining AF, probably contributing to worse recurrences of AF in patients undergoing a PVAI of AF. Thus, the physicians should be aware of the possibility of the presence of ECs even though the conventional PVAI is completed, when performing RFCA of AF.

Acknowledgments

We thank Mr Asami Yamada, Kensuke Kawasaki, Tomomi Hatae, Shu Takata, and Tsutomu Yoshinaga for their technical assistance with the electrophysiological study in the cardiac catheterization laboratory and Mr John Martin for his linguistic assistance with this paper.

References

- Haissaguerre M, Jais P, Shah DC, et al. Spontaneous initiation of atrial fibrillation by ectopic beats originating in the pulmonary veins. N Engl J Med 1998;339:659–666.
- Luik A, Radzewitz A, Kieser M, et al. Cryoballoon versus open irrigated radiofrequency ablation in patients with paroxysmal atrial fibrillation: the prospective, randomized, controlled, noninferiority FreezeAF Study. Circulation 2015; 132:1311–1319.
- Takahashi A, Iesaka Y, Takahashi Y, et al. Electrical connections between pulmonary veins: implication for ostial ablation of pulmonary veins in patients with paroxysmal atrial fibrillation. Circulation 2002;105:2998–3003.
- Cappato R, Negroni S, Pecora D, et al. Prospective assessment of late conduction recurrence across radiofrequency lesions producing electrical disconnection at the

649

pulmonary vein ostium in patients with atrial fibrillation. Circulation 2003; 108:1599-1604.

- Nyuta E, Takemoto M, Sakai T, et al. Epicardial connections after a conventional pulmonary vein antrum isolation in patients with atrial fibrillation. Circ J 2022; https://doi.org/10.1253/circj.CJ-22-0182.
- Barrio-Lopez MT, Sanchez-Quintana D, Garcia-Martinez J, et al. Epicardial connections involving pulmonary veins: the prevalence, predictors, and implications for ablation outcome. Circ Arrhythm Electrophysiol 2020;13:e007544.
- Niiyama D, Tsumagari Y, Uehara Y, Baba M, Hasebe H, Yoshida K. An epicardial connection with a unidirectional conduction property from the left atrium to pulmonary vein. JACC Case Rep 2022;4:310–314.
- Yoshida K, Baba M, Shinoda Y, et al. Epicardial connection between the rightsided pulmonary venous carina and the right atrium in patients with atrial fibrillation: a possible mechanism for preclusion of pulmonary vein isolation without carina ablation. Heart Rhythm 2019;16:671–678.
- Segerson NM, Lynch B, Mozes J, et al. High-density mapping and ablation of concealed low-voltage activity within pulmonary vein antra results in improved freedom from atrial fibrillation compared to pulmonary vein isolation alone. Heart Rhythm 2018;15:1158–1164.
- Chen S, Meng W, Sheng He D, et al. Blocking the pulmonary vein to left atrium conduction in addition to the entrance block enhances clinical efficacy in atrial fibrillation ablation. Pacing Clin Electrophysiol 2012;35:524–531.
- Willems S, Weiss C, Risius T, et al. Dissociated activity and pulmonary vein fibrillation following functional disconnection: impact for the arrhythmogenesis of focal atrial fibrillation. Pacing Clin Electrophysiol 2003; 26:1363–1370.