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A case of an atrial tachycardia originating from an occluded coronary sinus ostium with a persistent left superior vena cava

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ABSTRACT

We present a case of a 37-year-old female who complained of frequent palpitations caused by an atrial tachycardia and atrial premature contractions. Angiography revealed that the coronary sinus was occluded at the ostium and connected to a persistent left superior vena cava. An electrophysiological study and three-dimensional mapping revealed that the origin of the atrial tachycardia and atrial premature contractions was at the coronary sinus ostium in the right atrium. After repeat applications of radiofrequency energy at that site, no further atrial tachycardia or atrial premature contractions were induced by atrial burst pacing. To the best of our knowledge, this is the first report of an atrial tachycardia originating from an occluded coronary sinus ostium.

1. Introduction

Patients with an occlusion of the coronary sinus ostium with a persistent left superior vena cava, which is a rare cardiac anomaly, have been reported to suffer from atrial arrhythmias. In this article, we report a case associated with atrial premature contractions (APCs) and a focal atrial tachycardia (AT) originating from the occluded coronary sinus ostium.

2. Case report

A 37-year-old woman, previously healthy, visited a clinic complaining of frequent palpitations. Ambulatory electrocardiography (Holter ECG) revealed frequent APCs and paroxysmal AT with an uniform pattern coincident with palpitations. She was referred to our hospital for treatment of these arrhythmias. In the 24 hour Holter ECG, the P wave morphology of the APCs and non-sustained AT appeared similar and exhibited a negative polarity in both the CM5 and NASA leads (Fig. 1). Transthoracic echocardiography demonstrated almost normal findings except for mild mitral regurgitation, and the coronary sinus (CS) was not enlarged. After obtaining written informed consent, a cardiac electrophysiological study and RFCA were performed.

As no APCs appeared spontaneously during the procedure, we performed atrial burst pacing under an isoproterenol infusion, which provoked atrial fibrillation. After an electrical defibrillation, APCs and a non-sustained AT appeared. The P wave morphology of the APCs and AT was the same, with a negative polarity in leads II, III, and aVF, and positive polarity in lead aVL. Also, the P wave morphology was almost similar to that of the clinical APCs/AT. Multi-electrode catheters were placed in the superior vena cava, tricuspid valve annulus and His bundle area. We tried to insert a catheter into the CS but failed. Thereupon, we performed coronary angiography, which revealed that the ostium of the CS was occluded and that its distal end was connected to a persistent left superior vena cava (Fig. 2). Also, there were no coronary artery abnormalities like coronary artery fistulae observed in the angiography.

We attempted activation mapping of the APCs using a CARTO system (Biosense Webster, Diamond Bar, USA), but it could not be completed because the APCs did not appear very frequently. Instead, we kept mapping using a single catheter and could locate the site of the earliest activation of the APCs on the low right atrial septum. Eventually, an ablation catheter (Ablaze, Fantasista, 7Fr,

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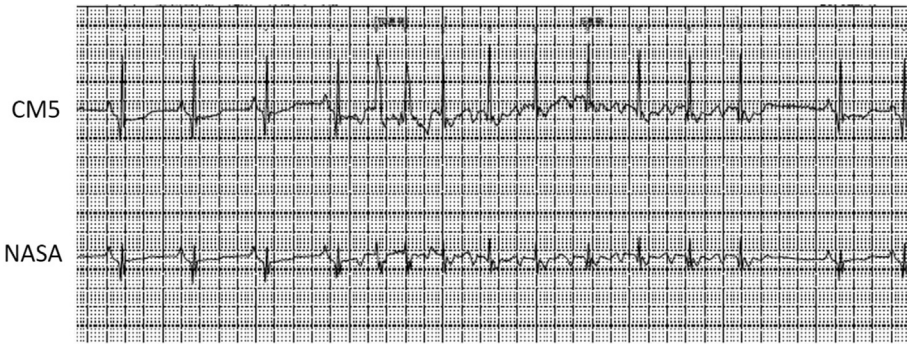

Fig. 1. In the 24 hour Holter ECG, a non-sustained atrial tachycardia appeared and its P wave had a negative polarity in both the CM5 and NASA leads.

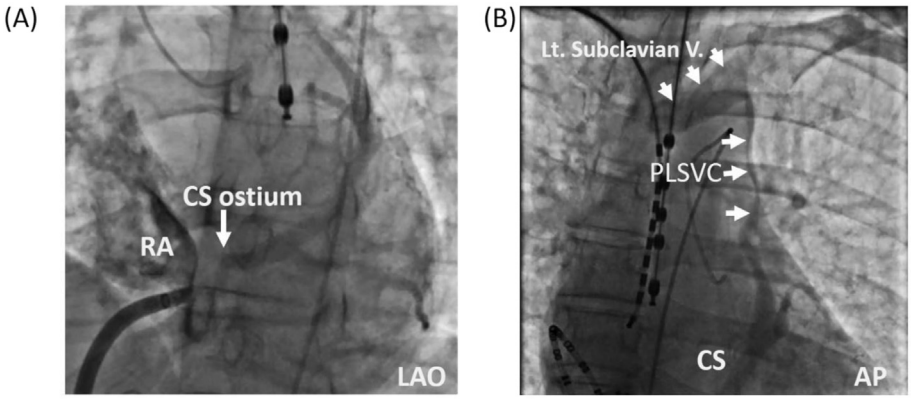

Fig. 2. (A) After a contrast injection into the left coronary artery (LCA), the right atrium was enhanced. A direct connection between the right atrium and coronary sinus could not be observed. (B) A persistent left superior vena cava appeared after a contrast injection into the LCA and it was connected to the left subclavian vein.
CS: coronary sinus, RA: right atrium, PLSVC: persistent left superior vena cava, LAO: left oblique view, AP: antero-posterior view.

Japan Lifeline Co., Ltd., Tokyo, Japan) was placed at the earliest activation site, where perfect pace mapping and a unipolar morphology with a QS pattern were achieved as shown in Figs. 3 and 4. The 1st RF energy application applied to that site succeeded to eliminate the APCs/AT followed by repetitive rapid atrial responses. After 20 additional RF applications in the vicinity of that site, no further arrhythmias could be provoked even with any type of atrial burst pacing under an isoproterenol infusion.

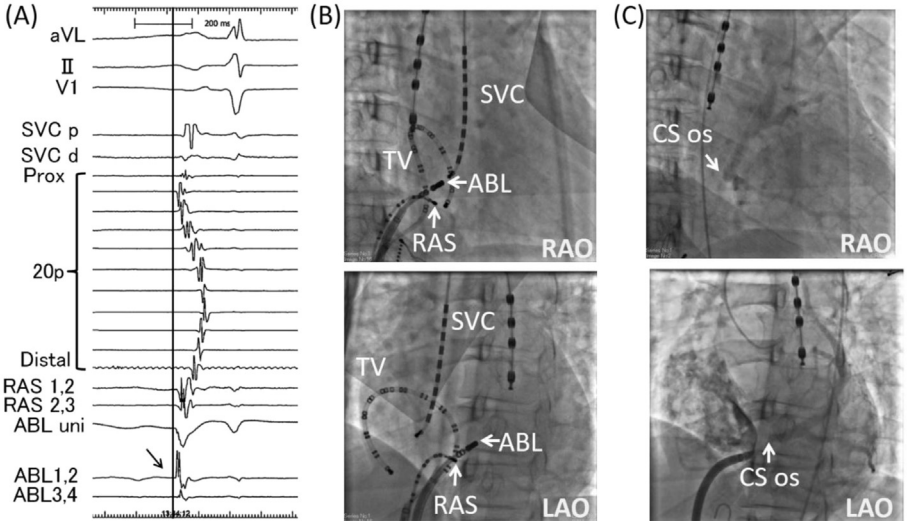

Fig. 3. The bipolar recordings at the successful ablation site exhibited the earliest activation site during an APC and the unipolar recording there exhibited a QS pattern. The successful ablation site was located in the right atrium, which was on the opposite side of the very proximal end of the coronary sinus.
RF: radiofrequency, SVC: superior vena cava, ABL: ablation, CS os: coronary sinus ostium, TV: tricuspid valve, HIS: His bundle, RAS: right atrial septum, RAO: right oblique view, LAO: left oblique view.

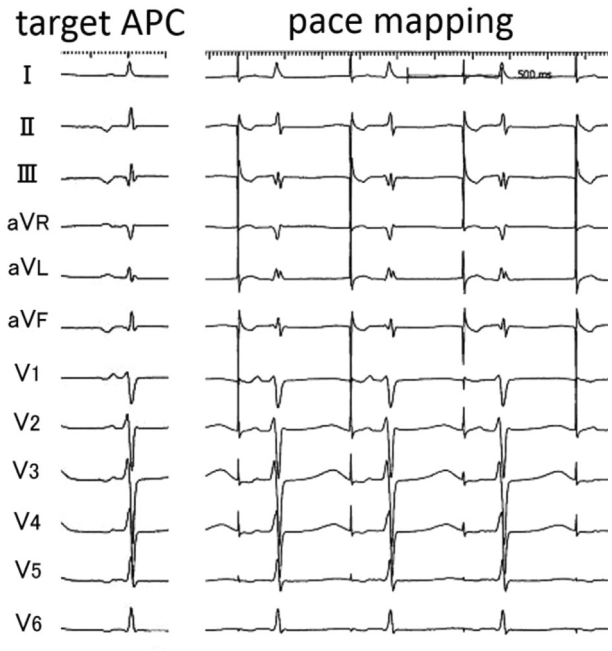

Fig. 4. Pace mapping of the P wave at the earliest activation site exhibited a perfect pace map.
APC: atrial premature contraction.

In the 24 hour Holter ECG after the ablation, the number of APCs decreased to 5 beats/day from 630 beats/day prior to ablation and no ATs were further recorded. During 50 months of follow-up, the patient has had no palpitations even on effort and repeated ECG recordings have revealed no APCs or ATs without any antiarrhythmic drugs.

3. Discussion

A CS ostial occlusion with a persistent left superior vena cava is usually associated with an anomalous connection between the coronary sinus and left atrium [1–3]. Since it is difficult to detect this anomaly in the clinical setting, it is usually reported in autopsy cases or during cardiac surgery, detected by an engorgement of cardiac veins and the heart turning bluish in color after a ligation of the persistent left superior vena cava. However, with the introduction of catheter ablation, this anomaly has been gradually recognized because it can be detected when the catheter cannot be advanced into the CS.

In our case, because the right atrial CS ostium was possibly congenitally occluded the blood from the coronary sinus traveled upward into the persistent left superior vena cava, and then into the right superior vena cava, by way of the left subclavian vein, and eventually into the right atrium. From an embryological point of view, the coronary sinus and oblique vein of Marshall are remnants of the vitelline, omphalomesenteric and common cardinal veins after they are occluded during embryonic weeks 5–10 from the left vein horn. If this occlusion of the left cardinal vein does not proceed normally, a persistent left superior vena cava is left.

Khairy et al. reported that an occluded coronary sinus ostium with a persistent left superior vena cava had been reported in only 40 patients up until 2005 [4]. There have only been 8 previously reported cases of arrhythmias associated with an occluded coronary sinus ostium and a persistent left superior vena cava [4–7]. The arrhythmias in those 8 cases included three with atrioventricular reentrant tachycardia (AVRT) and Wolff-Parkinson-White syndrome (type A: two, type B: one), two with atrioventricular nodal reentrant tachycardia, one with an AT from the superior aspect of the mitral annulus, two with common atrial flutter, and one with paroxysmal atrial fibrillation. None of those arrhythmias had an anatomically direct relationship to the occluded coronary sinus ostium. It is unknown whether the occluded coronary sinus ostium itself causes arrhythmias or not, and to the best of our knowledge, this is the first report of APCs/AT that have originated from an occluded CS ostium.

In 6 of the 8 reported cases, a diagnostic electrode catheter was placed in the CS via a persistent left superior vena cava, in which only one case with the type A WPW syndrome required recording with a CS catheter and none reported any difficulty of inserting the catheter into the persistent left superior vena cava. In our case, we could successfully locate and ablate the focus of the APCs/AT without a CS catheter recording, which meant that we did not try to place a CS catheter via the left superior vena cava. Instead, we determined the target site by activation mapping using bipolar and unipolar electrical potentials, and also by pace mapping. If the ablation was not successful from the right atrium or if one cannot use 3D mapping, it could be useful to place a CS catheter via the left superior vena cava. Because those APCs and non-sustained AT occurred spontaneously from a focal origin and were not provoked by atrial programmed pacing, the mechanism of those was speculated not to be reentry but automaticity or triggered activity. The 12-lead ECG was helpful for detecting the APC focus in our case and the evaluation of the P wave morphology of the AT recorded preablation was similarly useful for detecting the focus of the AT originating from the CS ostium in a previous report [8].

4. Conclusion

We report a case who had symptomatic atrial tachyarrhythmias associated with an occluded CS and persistent left superior vena cava, in whom catheter ablation was feasible for eliminating the arrhythmias by delivering RF applications to the occluded CS ostium.

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