A case of successful catheter ablation for biatrial reentrant tachycardia after a Mustard operation for dextro-transposition of the great arteries

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Introduction

Mustard and Senning operations are atrial switch techniques for dextro-transposition of the great arteries (d-TGA) that have been mainly performed during a period from the 1960s to the mid-1980s.

In the Mustard procedure, an artificial patch forming a baffle was used for the purpose of dividing the original atria into the following 2 chambers: a systemic venous atrium (SVA), which directs venous blood from the superior vena cava (SVC) and inferior vena cava to the mitral annulus and left ventricle; and a pulmonary venous atrium (PVA), which directs pulmonary blood to the tricuspid annulus and right ventricle.[1]

Despite survival improvement in these populations, complications such as right ventricular failure, arrhythmias, tricuspid valve dysfunction, and baffle-related problems over a long period are known.[2] In particular, incidence of supraventricular tachycardia (SVT) after the Mustard operation was reported as high as 48% owing to the extensive incisions and suture lines in the atria that resulted in intra-atrial conduction delays.[3]

Biatrial tachycardia (BiAT) is a rare form of macroreentrant atrial tachycardia (AT) that includes 2 independent interatrial connections and both atria within its circuit, and is often observed in patients with a history of catheter ablation for atrial fibrillation or open heart surgery.[4] Regarding congenital heart disease (CHD) patients, BiAT was reported to occur in approximately 4%; and a few cases after Senning operation were reported, but not in cases after Mustard operation.[5] We herein present a case of BiAT using upper and lower interatrial connections around the artificial patch after a Mustard operation.

KEY TEACHING POINTS

- Biatrial tachycardia after Mustard operation has not been reported before. The circuit involved both atria and upper and lower interatrial slow conductions around the artificial patch that separates systemic venous atrium and pulmonary venous atrium.
- Confirming postpacing interval – tachycardia cycle length $<$ 30 ms sites by entrainment pacing in multiple sites of both atria, as well as a biatrial activation map using a 3-dimensional mapping system, were mandatory to diagnose biatrial tachycardia.
- Understanding specific anatomy after Mustard operation and local potential features may lead to circuit identification.

Case report

A 41-year-old man who was diagnosed with d-TGA complicated with ventricular septal defect (VSD) underwent an atrial switch procedure by a Mustard operation combined with a patch closure for VSD at the age of 3 months. He also has a history of pulmonary venous pathway stenosis and bifemoral vein occlusion, which were identified in his teenage years. He underwent percutaneous transcatheter balloon angioplasty for pulmonary venous pathway stenosis at the age of 40.[6]

He developed repetitive palpitation and a 12-lead electrocardiogram showed narrow QRS tachycardia, which suggested AT. The echocardiogram demonstrated a mid-range

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right ventricular dysfunction with an ejection function of 40% that was responsible for systemic circulation, and a preserved left ventricular function with an ejection function of 53% that was responsible for pulmonary circulation. Because AT was symptomatic and intolerant to antiarrhythmic drugs, catheter ablation was performed under general anesthesia after written informed consent was obtained.

A decapolar catheter was placed in the left atrial appendage via the left subclavian vein as a reference catheter, which revealed 2-to-1 atrioventricular conduction AT with a cycle length of 290 ms. Owing to the obstruction of both femoral veins, the right jugular vein was selected as venous access to the SVA. An activation map of SVA performed by using a multipolar catheter and 3-dimensional mapping system (Pentaray, CARTO3; Biosense Webster, Inc, Irvine, CA) showed a focal pattern with the earliest activation site of the lower septum, which filled 79% of tachycardia cycle length (TCL) (Figure 1A).

Subsequently, the PVA activation map was performed using an irrigated-tip ablation catheter (ThermoCool SmartTouch; Biosense Webster, Inc, Irvine, CA) via a retrograde transaortic approach. The map showed a focal pattern with the earliest activation site of the ostium of the right superior pulmonary vein (RSPV), which filled 84% of TCL (Figure 1B and 1C). The earliest site of PVA structurally faced the SVC on the SVA side. The map of the left pulmonary vein (LPV) side, which is farther beyond the pulmonary venous pathway stenosis, was omitted because the local activation time on the LPV side during AT was well delayed enough to show a focal pattern of PVA with RSPV ostium as the earliest site.

Referring to voltage maps, a wide low-voltage area was found in the septum of both atria, suggesting an artificial patch.

Entrainment pacing studies showed that the site of "post-pacing interval (PPI) − TCL interval < 30 ms" was confirmed at the earliest site of SVA, the posterior SVC that was adjacent to the earliest site of PVA, the septum of PVA that faced the earliest site of SVA, and the posterolateral site of PVA (Figure 1). The "PPI−TCL < 30 ms" sites were located in discrete points in both atria.

The biatrial activation map covered the entire TCL. The wavefront commences from the anterior ostium of RSPV, descends through the posterolateral wall to the lower septum of the PVA, then enters the lower septum of the SVA, after which it ascends the SVA to the posterior SVC, and returns to the ostium of RSPV (Figure 2). The earliest activation site of each atrium faced the contralateral atrium chamber. Combined with the entrainment pacing findings, the AT was diagnosed as BiAT.

Notably, continuous fractionated potentials bridging double potential were recorded at the earliest site of each atrium (Figure 2, a and f), suggesting slow conduction pathways adjacent to the patch edge. The circuit consisted of the upper and lower interatrial conduction around the patch and both atria.

Radiofrequency (RF) applications to the earliest site of PVA resulted in TCL prolongation by 30 ms without atrial sequence change. The subsequent application to the earliest site of SVA prolonged TCL by 60 ms with a local potential delay by 100 ms without obvious atrial sequence change, though insertable electrode catheters were limited.

The PVA septum facing the earliest site of SVA was a wavefront entrance from PVA to SVA and was the subsequent target. While the Pentaray catheter was positioned at the lower septum of SVA as an anatomical marker, RF applications to the target where the local potential preceded the lower septal SVA via a retrograde transaortic approach successfully terminated BiAT.

Additional applications in this area were attempted but could not create the lower interatrial conduction block. Based on the SVA activation map during right atrial appendage pace on PVA, both upper and lower interatrial conductions remained; however, noninducibility of any ATs was achieved. The patient was free from any arrhythmia for more than 2 years without any antiarrhythmic drugs.

Discussion

SVT is commonly related to increased mortality and morbidity in patients with CHD.[7] SVT and impaired ventricular function are associated with risk factors of sudden cardiac death that have been reported at 2%–9% per decade.[8] Therefore, rhythm control is an important treatment in this population.

It was previously reported that in patients after atrial redirection procedure for TGA, the most common mechanism of SVT was cavotricuspid isthmus–dependent intra-atrial reentrant tachycardia (IART), followed by incisional or scar-related IART and focal AT.[9] In 12 cases of IART after Mustard operation, cavotricuspid isthmus was also reported to be the most frequent central barrier of IART, but there was no case of BiAT.[10]

Kitamura and colleagues[4] categorized BiAT into 3 types of reentrant circuits in cases with post-RF catheter ablation for atrial fibrillation (n = 7) and open heart surgery for an atrial septal defect (n = 1). Type 1 includes 2 separate interatrial connections and both atria, while types 2 and 3 include 2 separate interatrial connections and the right atrial septum.[4] In the present case, types 2 and 3 were excluded because the atrial septum was an artificial patch.

For BiAT series in patients with CHD, Moore and colleagues[5] reported 3 cases of BiAT after Senning operation. In the Senning procedure, self-atrial muscles are used to reconstruct SVA and PVA instead of using an artificial patch. Thereby the biological septa and complex suture lines may form substrates for diverse reentrant circuits, leading to several previously reported cases of BiAT.

To our knowledge, this is the first case report of BiAT after Mustard operation. Each atrial activation map demonstrated a focal pattern, and the biatrial activation map covered the entire TCL. By entrainment pacing, sites of PPI−TCL less than 30 ms were widely dispersed in both atria, including the earliest activation area of SVA and PVA. To diagnose the circuit as BiAT, entrainment pacing studies in both atria and biatrial

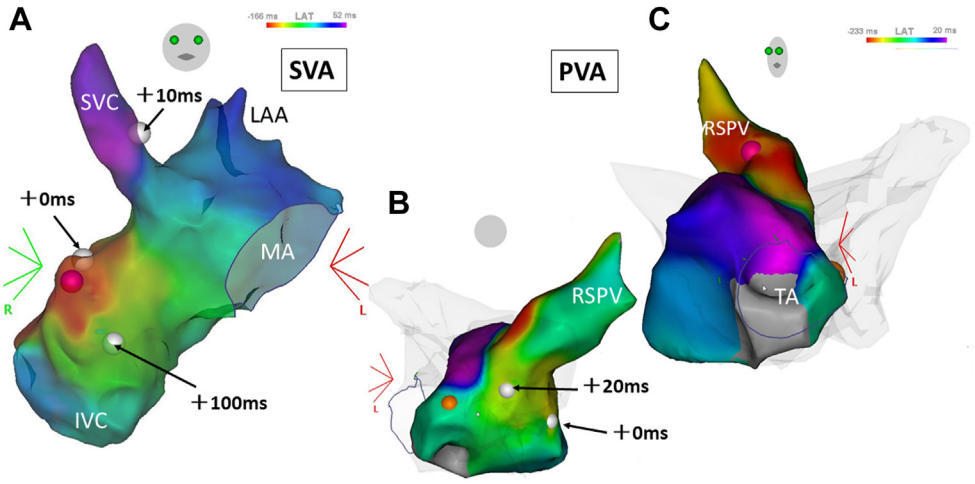

Figure 1 Activation map of each atrium and entrainment pacing studies. **A:** The activation map of systemic venous atrium (SVA) from the left anterior view shows a focal pattern with the earliest site of the lower posterior septum. **B, C:** The activation map of pulmonary venous atrium (PVA) from posterior view (**B**) and left anterior oblique view (**C**) show a focal pattern with the earliest site of right superior pulmonary vein (RSPV) ostium. Each earliest site faced the contralateral atrium chamber. The entrainment pacing sites are shown with white tags. "postpacing interval − tachycardia cycle length (TCL) < 30 ms" sites are located in discrete points in both atria. The entrainment pacing site at inferior vena cava (IVC) closest to the cavotricuspid isthmus in image A was out of the circuit. Pink tags in images A and C show effective ablation sites that resulted in TCL prolongation. The orange tag in image B shows a successful ablation site. LAA = left atrial appendage; MA = mitral annulus; SVC = superior vena cava; TA = tricuspid annulus.

activation map using a 3-dimensional mapping system were needed.

In this case, the local potential on the earliest site of each atrium showed continuous fractionated potentials bridging double potential. This electrical feature suggests a conduction delay on the suture line. There should be varying degrees of residual interatrial conduction between the 2 atria on the patch suture line, including conduction block, conduction delay, and conduction gaps. Based on the voltage map and local potentials, the patch area was estimated as shown in Figure 3. The circuit consisted of the upper interatrial connection that formed an entrance of excitation propagation from SVA to PVA (Figure 3A and 3B), the lower connection that formed an entrance from PVA to SVA (Figure 3C), and both atria.

RF applications to the earliest site of each atrium prolonged TCL. Especially, the ablation to the earliest site of SVA resulted in remarkable TCL prolongation and local excitation delay, which suggested that the lower interatrial connection was modified and changed to an alternative

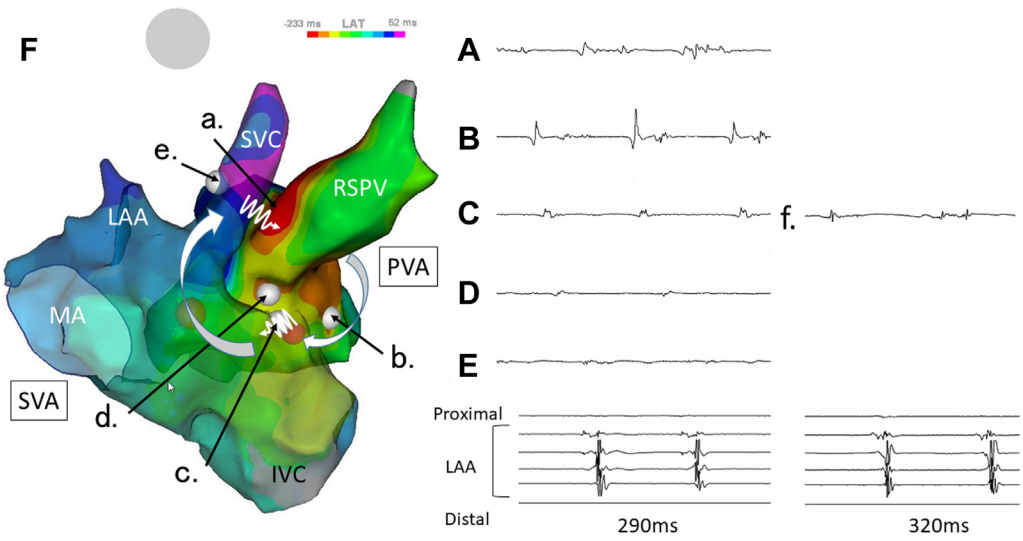

Figure 2 Biatrial activation map from posterior cranial view and local potentials at each location: **a:** the earliest activation site of pulmonary venous atrium (PVA); **b:** posterolateral wall of PVA; **c:** the earliest activation site of systemic venous atrium (SVA); **d:** posterior septum of PVA facing the earliest site of SVA; **e:** superior vena cava (SVC) adjacent to the earliest site of PVA; and **f:** the earliest site of SVA adjacent to site d after tachycardia cycle length (TCL) prolongation by 30 ms by ablation of site a. Postpacing interval − TCL < 30 ms was confirmed at sites b, c, d, and e. Continuous fractionated potentials between double potential were observed at the earliest sites of each atrial chamber, a and f. Other abbreviations as in Figure 1.

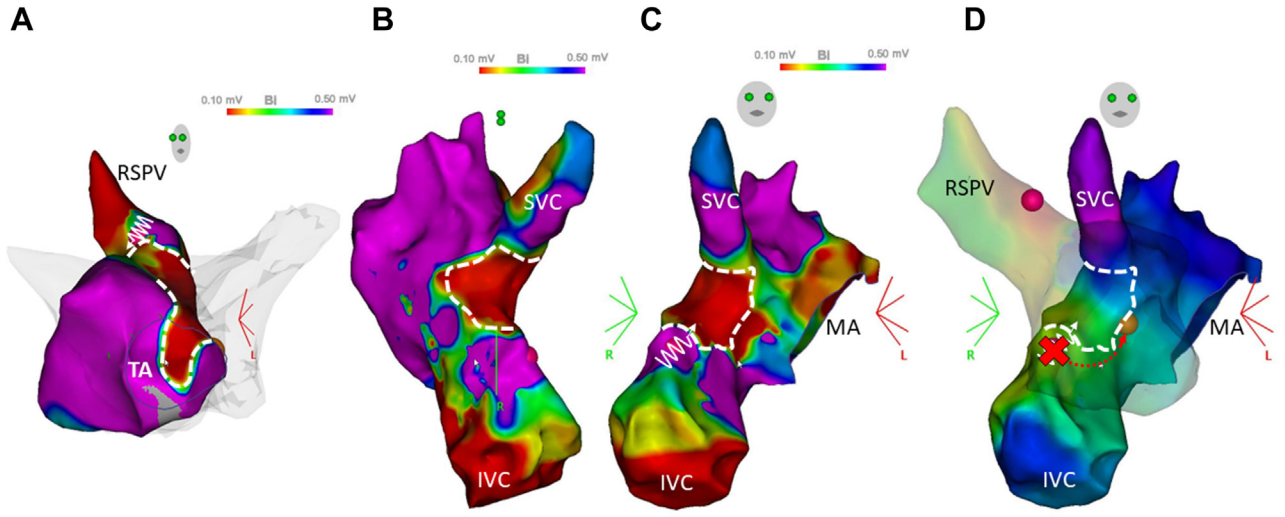

Figure 3 Estimation of septal patch location based on a voltage map and effective ablation sites. The septal patch edge is drawn with white dotted lines. **A:** A voltage map of pulmonary venous atrium (PVA) from left anterior oblique view shows that the entrance of excitation propagation from systemic venous atrium (SVA) to PVA is located on the upper edge of the patch (*white waved arrow*). **B:** A voltage map of SVA from right lateral view. The patch was sutured just below the superior vena cava (SVC) with its posterior edge adjacent to the right superior pulmonary vein (RSPV). **C:** A voltage map of SVA from right anterior oblique view shows that a propagation entrance from PVA to SVA is located on the lower edge of the patch that faced PVA (*white waved arrow*). **D:** Biatrial activation map from right anterior oblique view shows the successful ablation site in PVA with an orange tag. After effective ablation to the earliest site of SVA (*red cross mark*) resulted in tachycardia cycle length prolongation by 60 ms, the entrance was estimated to have changed from the lower edge to the anterior edge of the patch, which is an alternative pathway (*red dotted arrow*), because the success site of PVA was located anterior to the original earliest site of SVA 2 centimeters apart. Abbreviations and the descriptions of pink and orange tags are as in Figure 1. The voltage cut-off level was between 0.1 and 0.5 mV.

pathway in the entry site. The success site on PVA was the contralateral site of the earliest site of SVA, but it was located anterior to the original SVA earliest site, 2 cm apart. Because there could be multiple or wide interatrial connections around the patch, the entrance could have changed from the lower edge of the patch to the anterior one after RF applications to the inferior connection (Figure 3D).

Among the previously reported SVT cases after Mustard and Senning operations that were diagnosed as focal AT, there might have been underrecognized cases of BiAT because the activation pattern of 1-sided atrium presents a focal pattern. Even in such cases, it would be possible to treat by ablation to the earliest site of the atrium if the critical conduction could be eliminated without elucidating its entire circuit. However, careful electrophysiological mapping with recent advanced mapping systems may improve the identification of these complex circuits.

Considering the moderate recurrence rate of 10%–32% in these populations,[3,9,10] the present case has had a successful course. This probably supports that the critical conduction on the reentrant circuit was eliminated despite remaining interatrial connections.

Conclusion

We describe a rare case of BiAT involving upper and lower interatrial conductions around the artificial patch after Mustard operation. To diagnose, the entrainment pacing in both atria and biatrial activation maps were required. For effective ablation, biatrial approaches were also required.

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