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CASE REPORT
Arrhythmias/electrophysiology

Multiple peritricuspidal reentry tachycardias after cardiac infiltration by leukaemia: a case report

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Background Cardiac involvement by malignant lymphocytic neoplasms is a rare phenomenon. Little is known concerning cardiotoxicity in the chronic phase after completion of treatment.

Case summary A 50-year-old woman with a past history of cardiac involvement of acute lymphoblastic leukaemia (ALL) underwent electrophysiologic study and catheter ablation for symptomatic atrial tachycardia (AT). She was diagnosed with ALL when she was 8 years old and treated with systematic chemotherapy with prednisolone and vincristine. After complete remission, she suffered from repeated palpitations beginning at the age of 16 years. Electrophysiologic study using high-density (HD) mapping showed two types of peritricuspid AT in addition to low voltage in the right atrium with conduction delay.

Discussion Cardiac involvement by malignant lymphocytic neoplasms is a rare phenomenon, and cardiac infiltration often disappears after remission of ALL. Thus, little is known about cardiac electrophysiological characteristics in the chronic phase of complete remission of ALL. We describe a rare case of a patient with multiple peritricuspidal reentry tachycardias after cardiac infiltration by leukaemia using a HD mapping system.

Keywords Macro reentry • Atrial flutter • Rhythmia mapping system • Leukaemia • Infiltration • Case report

Learning points

- After complete remission of acute lymphoblastic leukaemia, there is the possibility of an existing substrate for macroreentrant atrial tachycardia.
- Ablation for the isthmus between the inferior vena cava and tricuspid annulus may carry the risk of inducing intra-atrial conduction block in patients who have conduction delay in the right atrium. High-density mapping is a useful tool to create a safety strategy for ablation.

Introduction

Cardiac involvement by malignant lymphocytic neoplasms is a rare phenomenon, and has been reported in approximately 8.7–37% of autopsy cases involving lymphoma or leukaemia.[1] Cardiac infiltration often disappears after remission of acute lymphoblastic leukaemia (ALL).[2,3] With the exception of cardiotoxicity associated with chemotherapy (e.g. anthracyclines, doxorubicin, and daunorubicin) or radiation therapy, little is known concerning cardiotoxicity in the chronic phase after completion of treatment.[4] Macroreentrant atrial

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tachycardias (ATs) can occur in patients with organic heart disease or after cardiac surgery or catheter ablation of atrial fibrillation.

Timeline

8 years of age	Diagnosis of acute lymphoblastic leukaemia with cardiac infiltration. Systemic chemotherapy with prednisolone and vincristine. Achieved complete remission.
16 years of age	Symptomatic atrial tachycardia (AT) was treated by anti-tachycardia pacing.
41 years of age	AT was documented again. β-blockers were prescribed.
50 years of age	Electrophysiological study and catheter ablation was performed.

Case presentation

A 50-year-old woman presented to our hospital complaining of palpitation and dizziness during exercise. She had a history of ALL diagnosed at the age of 8 years. At initial diagnosis, cardiac infiltration by ALL with pericardial effusion was demonstrated by transthoracic echocardiography (TTE), and AT was documented. After systemic chemotherapy with prednisolone and vincristine, she achieved complete remission and the cardiac infiltration completely resolved. No arrhythmic event was observed until she was 16 years old. At that time, she developed sudden palpitations after exertion. Electrocardiography (ECG) demonstrated AT with a cycle length of 285 ms. Atrial tachycardia was terminated with anti-tachycardia pacing from the right atrium (RA) during electrophysiological study, but catheter ablation was not conducted. Atrial tachycardia was documented again when she was 41 years old, and β-blockers were subsequently prescribed for her symptoms. She had no other past medical history. She presented to our hospital to undergo catheter ablation for AT. Cardiovascular and respiratory examinations were unremarkable. Electrocardiography showed AT with a tachycardia cycle length of 340 ms; the P-wave configuration was negative in the inferior leads and the V1 lead (*Figure 1*). TTE showed normal systolic function without any cardiac infiltration or pericardial effusion. An electrophysiological study was performed after the patient provided informed consent.

An activation map of 15 471 points was acquired in 20.1 min in the RA during AT with the Orion multipolar basket catheter (Boston Scientific, Marlborough, MA, USA) and Rhythmia Mapping System (Boston Scientific). The activation map showed peritricuspid AT with a counter-clockwise activation pattern (*Figure 2* and Supplementary material online, *Video S1*). The first impression was that of common atrial flutter. However, the root of the RA appendage and upper RA

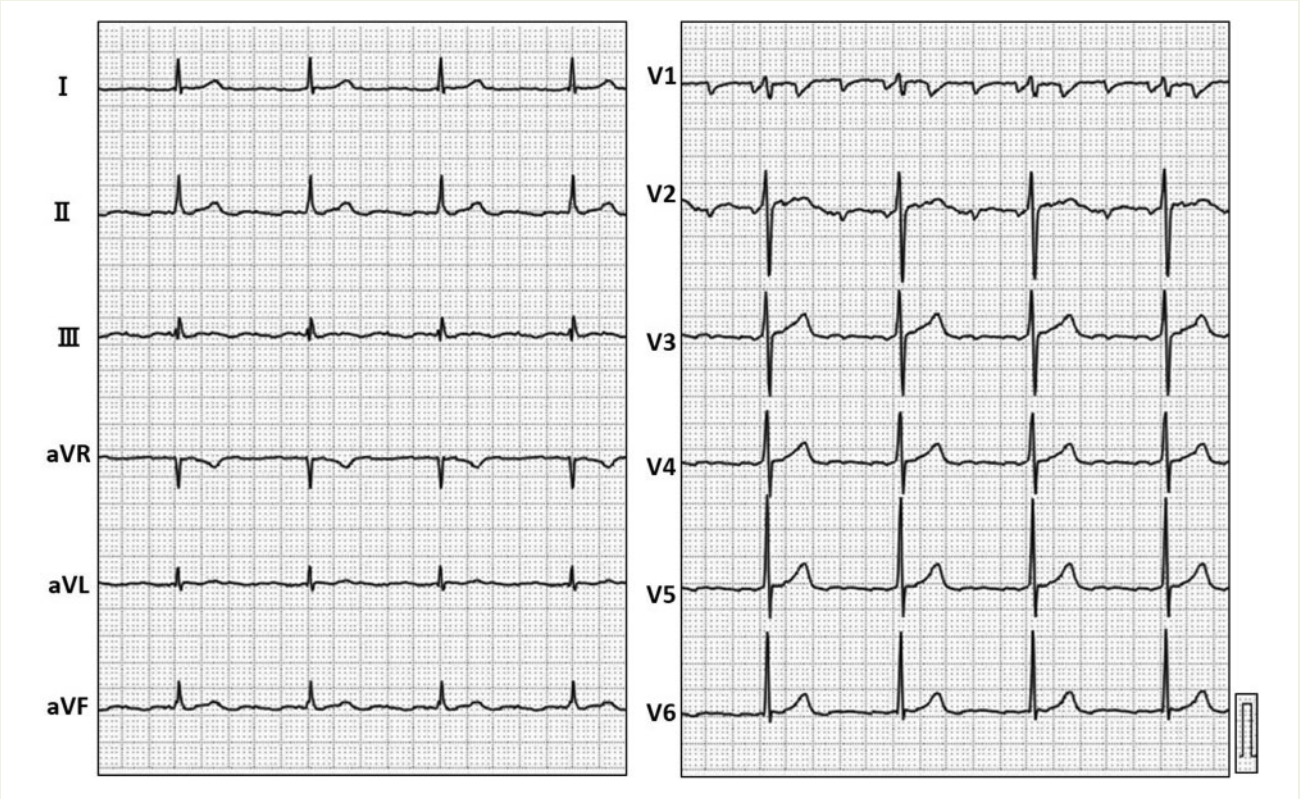

Figure 1 Twelve lead electrocardiogram. Electrocardiogram showed atrial tachycardia with a tachycardia cycle length of 340 ms; the P-wave configuration was negative in the inferior leads and the V1 lead.

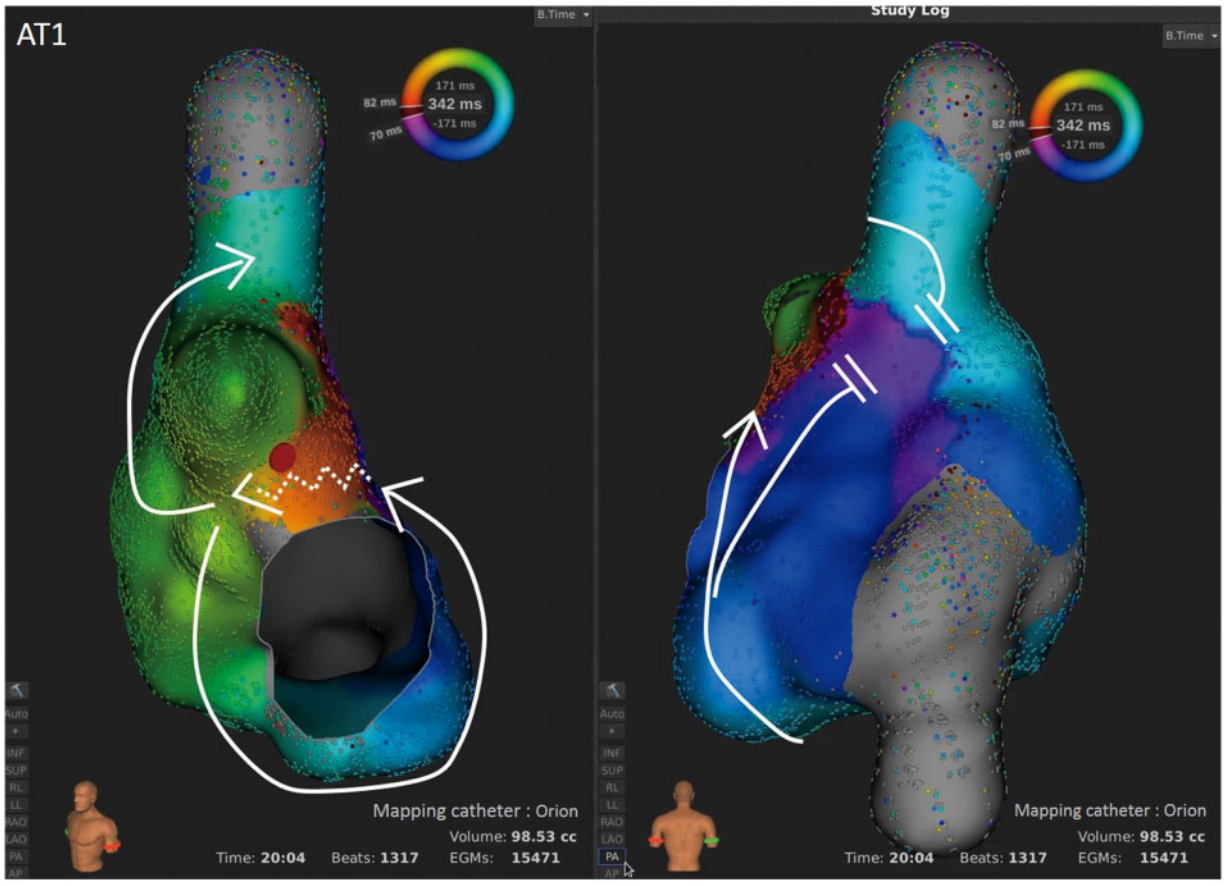

Figure 2 Activation map of atrial tachycardia 1. The activation map of atrial tachycardia 1 showed peritricuspid atrial tachycardia with a counter-clockwise activation pattern. Atrial tachycardia 1 was terminated by radiofrequency application to the root of the right atrial appendage (red tag).

septum showed conduction delays. Thus, ablation to the isthmus between the inferior vena cava and tricuspid annulus (TA) carried the risk of inducing intra-atrial conduction block. Avoiding isthmus ablation, catheter ablation was applied to the root of the RA appendage and AT was terminated during the ablation. After termination of the AT, RA mapping was performed under high RA pacing because the sinus node was suppressed. The voltage map during high RA pacing with 4896 points in 16.3 min demonstrated a low voltage area around the TA, including the root of the RA appendage, upper RA septum, and lateral free wall (*Figure 3*). The activation map showed upper RA septum conduction delay in the low-voltage area. The ablated area of the root of the RA appendage showed conduction block (Supplementary material online, *Video S2*). After RA mapping, sinus rhythm was recovered with 1 μg of isoproterenol (ISP) infusion, but the atrioventricular (AV) conduction time was prolonged at 320 ms. AT2 occurred spontaneously after mapping. An activation map with 8508 points in 12.8 min showed that AT2 was also a peritricuspid AT involving the upper septum slow conduction area (*Figure 4* and Supplementary material online, *Video S3*). Entrainment pacing from the upper septum was attempted; however, pacing was not able to capture the potentials stably due to the low voltage. AT2 was terminated during radiofrequency application to the upper RA septum. After linear ablation at the upper RA septum, tachyarrhythmia was not induced by programmed atrial stimulation and AV conduction was preserved. ISP was administered until the day after the procedure for sinus bradycardia. However, sinus node function recovered spontaneously, and she was discharged without prolonged hospitalization. After discharge, she was followed up in our outpatient clinic 1 and 3 months after the procedure and every 3 months thereafter. She was asked about her symptoms at each visit, and 24 h ambulatory Holter ECG and routine 12-lead surface ECG were carried out at 1, 3, 6, and 12 months and every 6 months thereafter. She has been free from any recurrence of tachyarrhythmia or bradycardia event requiring pacemaker for 18 months after the procedure.

Discussion

It has been reported that leukaemic infiltration is frequently identified on post-mortem examination of the myocardium and pericardium.[1] However, during ante-mortem examination, pericardial involvement is rare and after remission of leukaemia, cardiac infiltration usually disappears.[2,3] Thus, little is known concerning the cardiac electrophysiological characteristics in the chronic phase of complete remission of ALL. In this case, AT was detected when the patient was 16 years old

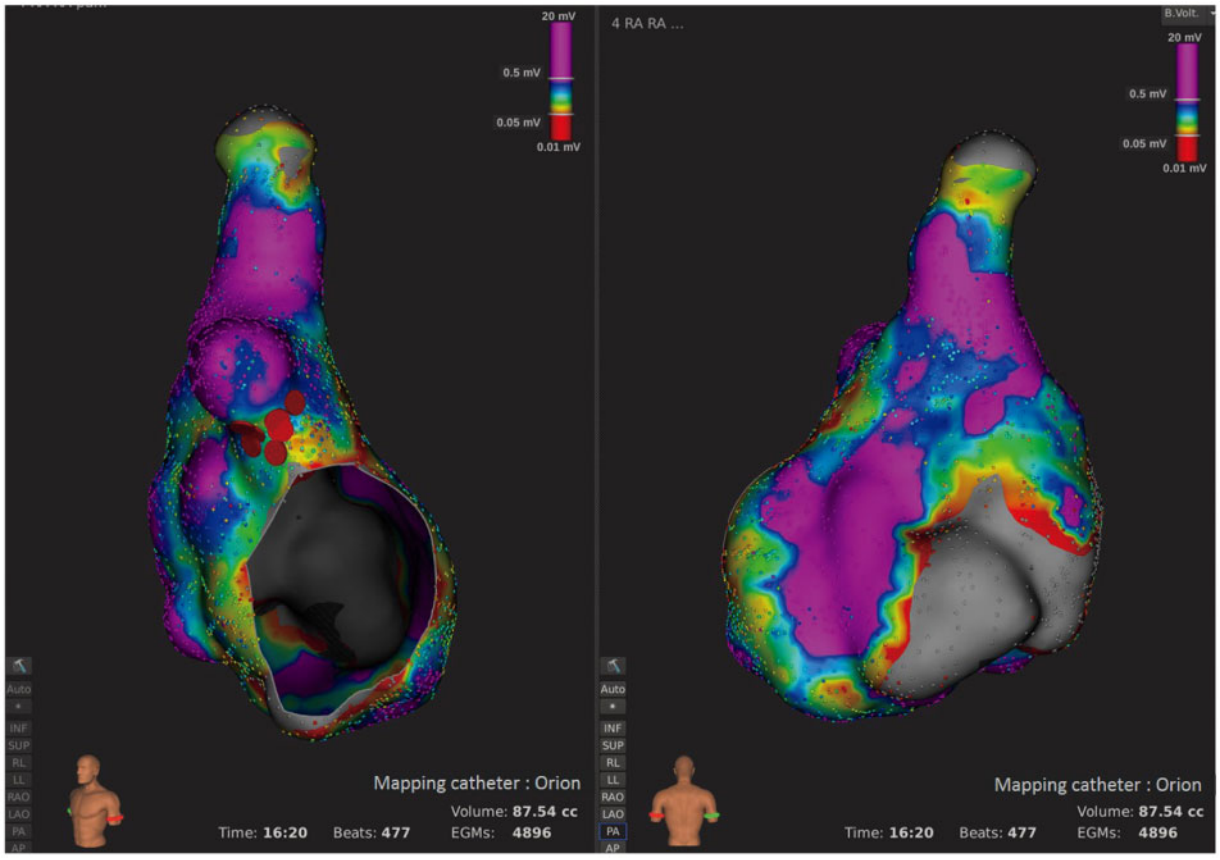

Figure 3 Voltage map during high right atrium pacing. The voltage map during high right atrium pacing demonstrated the low-voltage area around the tricuspid annulus including the root of the right atrium appendage, upper septum, and lateral free wall.

and continued until she was 41 years old. She was free from relapse at both of these time points. She had no prior history of surgery or cardiac disease with the exception of ALL. The macroreentrant AT was thought to be due to damage secondary to cardiac infiltration. There are a few reported cases of AT after resolution of cardiac involvement by ALL, but there is no report of detailed electrophysiological study after complete remission of ALL in the chronic phase.[4] We demonstrated the precise macroreentrant circuits and voltage map of the RA. This case implies that there is a possible substrate for macroreentrant AT even after complete remission of ALL. The high-density (HD) mapping system allowed us to create a detailed activation map and a slow conduction zone. Both types of ATs were peritricuspid AT, which is similar to common atrial flutter. However, ablation for the isthmus between the inferior vena cava and TA carried the risk of inducing an intra-atrial conduction block. Both ATs were successfully treated by radiofrequency application for slow conduction zones, which were detected by the HD mapping system. This approach allowed the preservation of intra-atrial conduction. One limitation of this case report is that we did not investigate the precise AV conduction time before ablation because we ablated the slow conduction zone during the ATs. After recognizing that there are zones of very slow conduction especially in the septal RA, it would be better to consider terminating the tachycardia and looking at AV conduction before the linear ablation. In conclusion, we report a rare case of macroreentrant AT in a patient after complete remission from ALL during the chronic phase. We should consider the possibility of myocardial damage, which may cause arrhythmia even after complete remission from ALL.

Lead author biography

Iwanari Kawamura, M.D. graduated from Hamamatsu University School of Medicine, Shizuoka, Japan in 2011. Resident in Tokyo Metropolitan Hiroo Hospital, Japan from 2011 to 2012. Fellow in Cardiology, Tokyo Metropolitan Hiroo Hospital, Tokyo, Japan from 2013 to 2016. Clinical Fellow in Arrhythmia and Electrophysiology, Tokyo Metropolitan Hiroo Hospital from 2017 to 2018. He was an Exchange program student, Department of Cardiovascular Surgery, Faculty of Medicine Siriraj Hospital, University of Mahidol, Bangkok, Thailand in 2010. Certification: Board Certified Member of the Japanese Society of Internal Medicine, Fellow of the Japanese Society of Internal Medicine (FJSIM), Board of

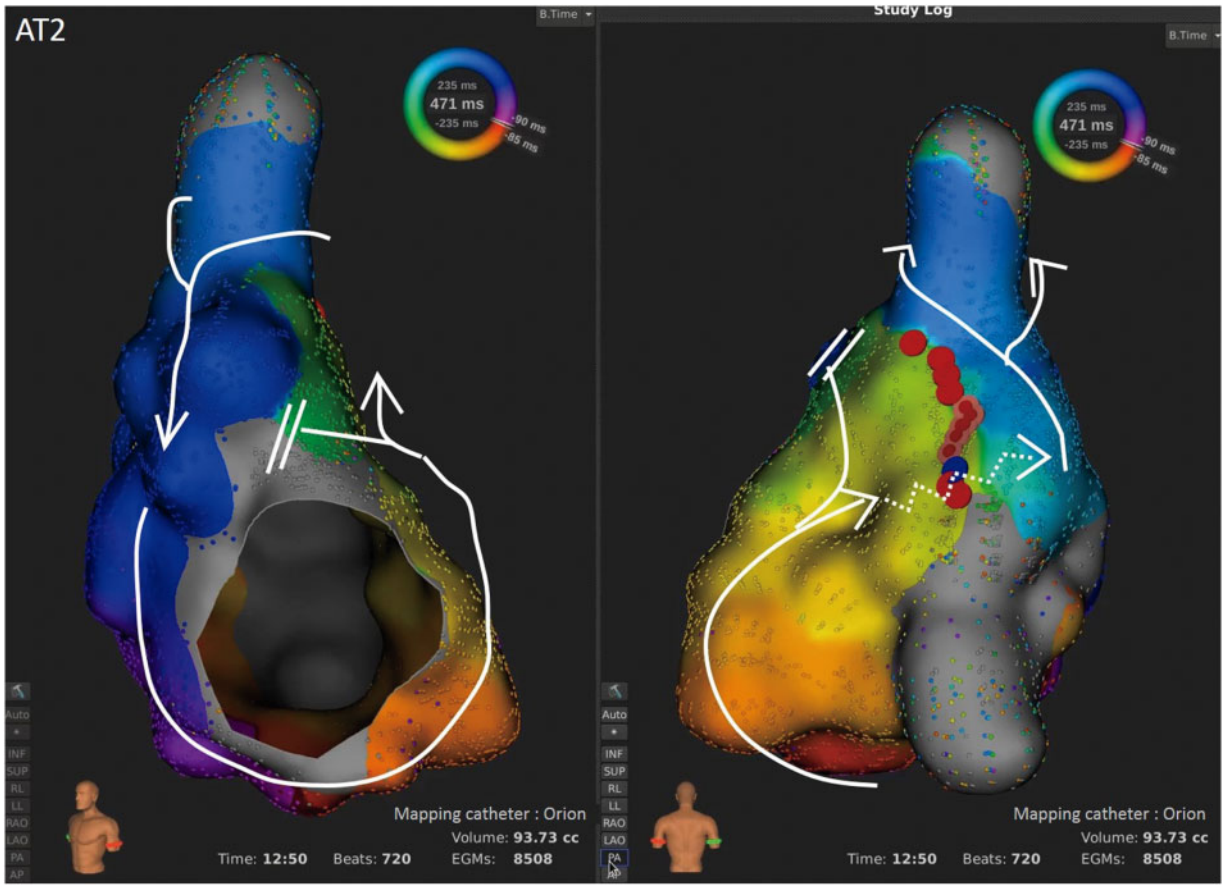

Figure 4 Activation map of atrial tachycardia 2. The activation map of atrial tachycardia 2 also showed peritricuspid atrial tachycardia with a counter-clockwise activation pattern. Atrial tachycardia 2 was terminated by radiofrequency application to the upper septum (blue tag). After termination of atrial tachycardia 2, an additional linear ablation was performed (red tag).

Japanese Association of Cardiovascular Intervention and Therapeutics. He is a Clinical fellow in Tokyo Metropolitan Hiroo Hospital. Membership: 2012 The Japanese Society of Internal Medicine, 2012 The Japanese Circulation Society, 2013 The Japanese Association of Cardiovascular Intervention and Therapeutics, 2013 The Japanese Heart Rhythm Society.

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: The author/s confirm that written consent for submission and publication of this case report including image(s) and associated text has been obtained from the patient in line with COPE guidance.

Conflict of interest: none declared.

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