



## Case Report

## Complex regional pain syndrome induced by pacemaker implantation for sick sinus syndrome

Megumi Kisanuki<sup>a</sup>, Kazumasa Fujita<sup>a</sup>, Shohei Moriyama<sup>a</sup>, Kei Irie<sup>a</sup>, Chiharu Yosida<sup>a</sup>, Mitsuhiro Fukata<sup>a</sup>, Takeshi Arita<sup>a</sup>, Taku Yokoyama<sup>a</sup>, Keita Odashiro<sup>a</sup>, Toru Maruyama<sup>b,\*</sup>, Koichi Akashi<sup>a</sup><sup>a</sup> Heart Center, Kyushu University Hospital, Maidashi 3-1-1, Fukuoka 812-8582, Japan<sup>b</sup> Faculty of Arts and Science, Kyushu University, Kasuga Kohen 6-1, Kasuga 816-8580, Japan

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## ABSTRACT

A 53-year-old woman reported burning pain, muscle weakness, and dysesthesia of the left arm 2 months after permanent pacemaker insertion in the ipsilateral side for the treatment of sick sinus syndrome. Complex regional pain syndrome (CRPS) induced by pacemaker implantation was diagnosed. In 2017, her pulse generator became exhausted and was exchanged carefully to avoid exacerbation of CRPS, under the application of local anesthesia and premedication. Six months later, the patient's grip strength in her left hand remained lower relative to that in her right hand. Although rare, the presence of CRPS following device implantation should be remembered.

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## 1. Introduction

Complex regional pain syndrome (CRPS) is conventionally known as reflex sympathetic dystrophy, and is characterized by an abnormal sensation of pain. It is classified either as CRPS without peripheral nerve injury (type 1) or as CRPS associated with nerve injury (type 2) [1]. This syndrome can be induced by minor trauma, including via medical procedures such as venipuncture. Iatrogenic CRPS after permanent pacemaker implantation is extremely rare [2–4]. We experienced a case of type 1 CRPS induced by permanent pacemaker implantation for sick sinus syndrome (SSS). When care was taken during the exchange of the expired pulse generator after recognition of this rare syndrome, CRPS did not exacerbate. Procedural local pain control is essential to prevent CRPS.

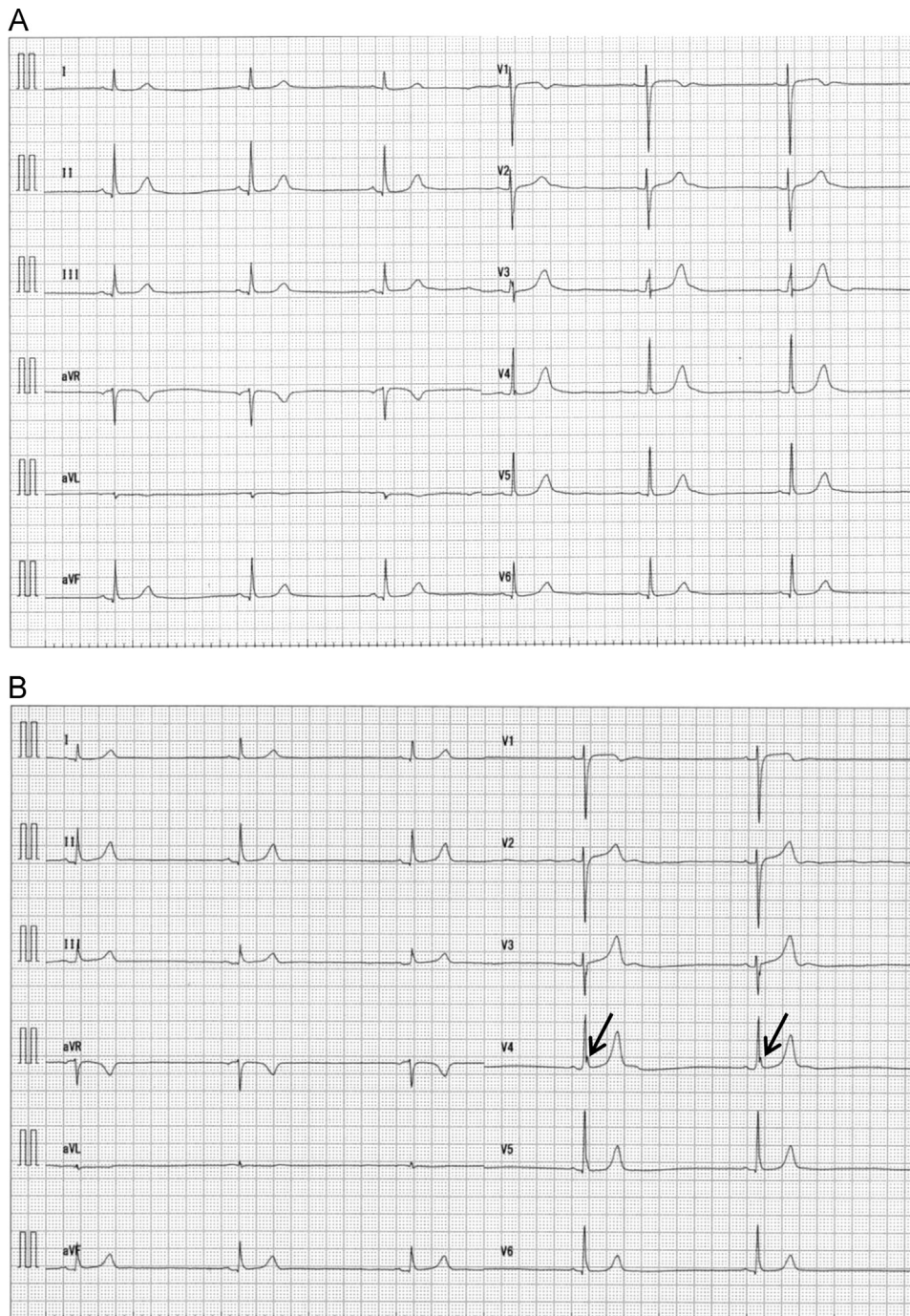
## 2. Case history

In August 2007, a 53-year-old woman complained of fatigue and dizziness and visited Kyushu University Hospital for the treatment of SSS, which was diagnosed by electrocardiogram (ECG) at her first visit (Fig. 1A). During the second visit one month later, another ECG showed similar findings. Therefore, cilostazol (200 mg/day) was administered in September 2007 to increase her heart rate (HR) [5]. An ECG recorded on admission in October 2007 showed no increase of HR (Fig. 1B). Echocardiography showed no organic heart diseases. Systemic diseases causing sinus bradycardia (such as hypothyroidism or elevation of intracranial pressure) were not found. The maximum RR interval was 4.9 s, and the minimum HR was 28 bpm. Electrophysiological study (EPS) to evaluate sinus node function was not performed, because SSS-induced symptoms were progressive and permanent pacemaker implantation appeared urgent. In October 2007, a permanent pacemaker (Identity DR<sup>TM</sup>, DDDR; St. Jude Medical, St. Paul, MN, USA) was implanted successfully without premedication under local anesthesia with 1% procaine alone. The patient complained of local pain after the procedure despite postoperative routine administration of nonsteroidal anti-inflammatory drugs (NSAIDs). Additional NSAIDs were prescribed, and she was discharged without any complications, such as residual local pain or left arm edema.

In January 2008, she complained of left arm swelling and pain followed by muscle weakness. These symptoms became worse until she could no longer make a fist with her left hand due to

\* Correspondence to: Faculty of Arts and Science, Kasuga-Kohen 3-1, Kasuga City, Fukuoka 816-8580, Japan. Fax: +81 92 592 2866.

E-mail addresses: [kisanuki.megumi@gmail.com](mailto:kisanuki.megumi@gmail.com) (M. Kisanuki), [mfukata-circ@umin.net](mailto:mfukata-circ@umin.net) (K. Fujita), [morilyn.monroe@gmail.com](mailto:morilyn.monroe@gmail.com) (S. Moriyama), [qxrng328@yahoo.co.jp](mailto:qxrng328@yahoo.co.jp) (K. Irie), [asunifaasuno@yahoo.co.jp](mailto:asunifaasuno@yahoo.co.jp) (C. Yosida), [mfukata-circ@umin.net](mailto:mfukata-circ@umin.net) (M. Fukata), [takeshi.arita@gmail.com](mailto:takeshi.arita@gmail.com) (T. Arita), [dreammaker1005@hotmail.com](mailto:dreammaker1005@hotmail.com) (T. Yokoyama), [odashiro@med.kyushu-u.ac.jp](mailto:odashiro@med.kyushu-u.ac.jp) (K. Odashiro), [maruyama@artsci.kyushu-u.ac.jp](mailto:maruyama@artsci.kyushu-u.ac.jp) (T. Maruyama), [Akashi@intmed1.med.kyushu-u.ac.jp](mailto:Akashi@intmed1.med.kyushu-u.ac.jp) (K. Akashi).



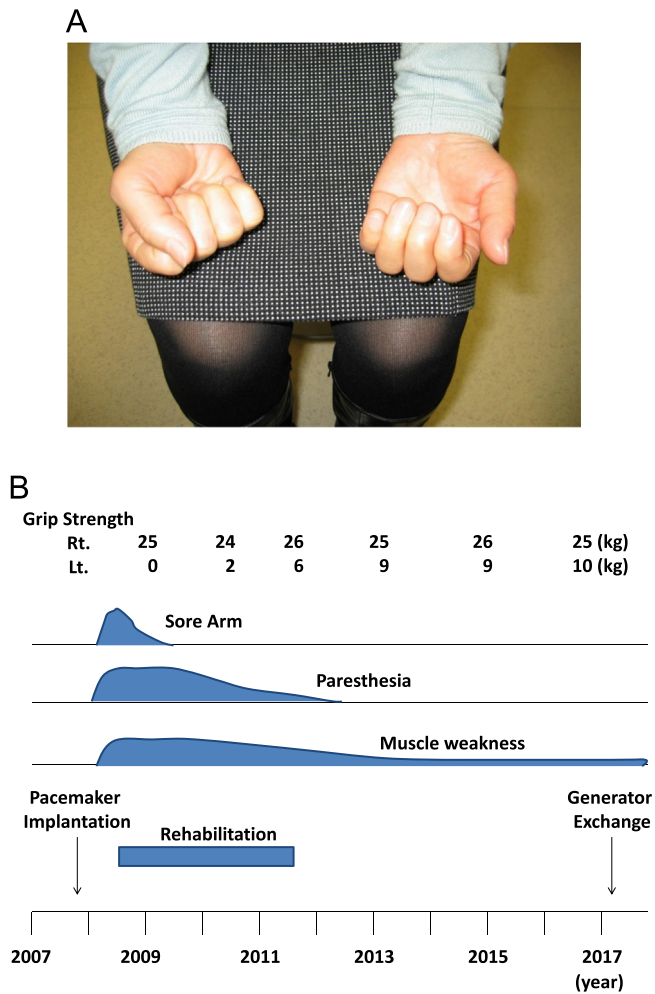
**Fig. 1.** ECGs recorded in August (A) and October (B) 2007, respectively. HR was 38 bpm at the first visit (A). Despite the administration of cilostazol (200 mg/day), the patient's sinus bradycardia did not improve (30 bpm). Note the bradycardia-dependent J waves (arrows) present in (B).

muscle weakness, finger stiffness, and skin tightness (Fig. 2A). Full-body computed tomography, gallium scintigraphy, and fluorodeoxyglucose positron emission tomography showed no specific abnormalities. A diagnosis of type 1 CRPS was made according to the current guidelines [1], after ruling out amyotrophic lateral sclerosis, myasthenia gravis, and sarcoidosis. Treatment with steroids or neurotrophin was recommended [2,4]. However, she was reluctant to start drug therapy. Therefore, she was encouraged instead to perform rehabilitation, focusing on the left arm; subsequently, the grip strength of her left hand gradually recovered (Fig. 2B). In January 2017, a regular pacemaker check revealed that the pulse generator had become exhausted. The pulse generator was exchanged carefully for an Assurity™ DR2240 (St. Jude

Medical, St. Paul, MN, USA) under premedication with dexmedetomidine followed by pentazocine and hydroxyzine and local anesthesia with xylocaine. Adequate pain control was ensured. Although CRPS was not exacerbated during this procedure, grip strength in her left hand remains lower relative to that in her right hand.

### 3. Discussion

We presented a patient with CRPS following permanent pacemaker implantation for progressive, drug-refractory SSS. CRPS is a rare clinical entity characterized by burning pain and swelling



**Fig. 2.** Photograph (A) of the patient's left and right hands (taken in 2011) and clinical time course (B).

followed by muscle atrophy, skin tightness, and joint stiffness [1]. Its induction by permanent pacemaker implantation is rare, and all known patients that have experienced CRPS following pacemaker implantation were elderly [2–4]. None of them had infection, pacemaker malfunction, or lead troubles. Accordingly, the predictors of post-implantation CRPS remain unclear.

Our patient was originally sensitive to pain, and permanent pacemaker implantation was her first experience of intervention, because she did not undergo invasive EPS. Although written informed consent was obtained, her mental preparedness may not have been at an adequate level prior to pacemaker implantation. Furthermore, unlike the other patients reported [2–4], the pulse generator was exchanged in our patient. The difference between the first implantation and the second procedure was the higher degree of attention paid in order to control the local pain so as to avoid the exacerbation of CRPS. The outcome suggests that local pain control during the procedure is essential to prevent CRPS in the elderly.

#### 4. Conclusions

We presented the case of a patient with CRPS induced by implantation of a permanent pacemaker. Pulse generator exchange was performed with recognition of the risk of the exacerbation of CRPS, and thus, adequate pain control was ensured.

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We have no sources of funding to disclose.

#### Conflict of interest

All authors declare no conflicts of interest related to this study.

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