

A case of twiddler's syndrome with a subcutaneous implantable cardioverter-defibrillator



Chengyue Jin, MD,* Sei Iwai, MD, FHRS,[†] Jason Jacobson, MD, FHRS,[†]
Aileen Ferrick, PhD, ACNP, FHRS[†]

From the *Department of Medicine, Westchester Medical Center, Valhalla, New York, and [†]Department of Cardiology, Westchester Medical Center, Valhalla, New York.

Introduction

Twiddler's syndrome (TS) is twisting of the pulse generator in a cardiac implantable electronic device (CIED) pocket, either deliberately or unintentionally, resulting in lead displacement and device malfunction.¹ TS with transvenous (TV) CIEDs has been well described, but there is limited data with subcutaneous implantable cardioverter-defibrillators (S-ICDs).

Case report

A 50-year-old male patient with hypertrophic cardiomyopathy and indication for primary prevention ICD underwent S-ICD implantation. The pulse generator was secured to the fascia with 2 0 silk sutures. Two nonabsorbable, braided sutures were placed around the suture sleeve near the subxiphoid, with the lead tunneled up the sternum. Two weeks post implant, the S-ICD demonstrated appropriate function. Two weeks later, remote monitoring reported 1 ICD shock (Figure 1A). Interrogation demonstrated reduced R waves and shock impedance. There were no reported preshock symptoms; the patient was clapping his hands during the occurrence. Chest radiography revealed the lead dislodged, retracted, and coiled around the generator in the left lateral chest wall (Figure 1B and 1C) as compared to implant chest radiograph (Figure 1D and 1E). The patient underwent lead revision and device replacement, and a 0 silk suture was placed at the tip of the lead to prevent future dislodgment.

Discussion

Incidence of TS is rare, ranging from 0.07% to 2.69%.^{2,3} An important risk factor is the patient manually manipulating the CIED.⁴ Advanced/younger ages, redundant skin, obesity, oversized pocket, and psychiatric disorders are other risk factors.⁵⁻⁷ Compared to TV-ICD, the S-ICD has lower rates of

early lead displacement (1% vs 2.7%), and lead malfunction (0% vs 6.2%).⁸ TS can be fatal by ventricular tachyarrhythmias being undetected or unsuccessfully treated. Work-up, if suspected, includes electrocardiogram, chest radiograph, and device interrogation.⁶ Studies show certain surgical techniques can reduce TS in TV-ICD, including anchoring sutures to the pectoralis fascia, as well as antimicrobial and woven Dacron pouches.⁹⁻¹¹ Patient education can be helpful but is not completely effective, given subconscious manipulation or spontaneous migration of devices.⁶ It is reasonable to infer that these strategies could also reduce risk of S-ICD TS.

Conclusion

Not unlike transvenous CIEDs, TS occurs in S-ICDs, with similar risk factors and preventive strategies.

References

1. Bayliss CE, Beanlands DS, Baird RJ. The pacemaker-twiddler's syndrome: a new complication of implantable transvenous pacemakers. *Can Med Assoc J* 1968; 99:371-373.
2. Fahraeus T, Hoijer CJ. Early pacemaker twiddler syndrome. *Europace* 2003; 5:279-281.
3. Kwon CH, Choi JH, Kim J, et al. Complications of cardiac perforation and lead dislodgement with an MRI-conditional pacing lead: a Korean multi-center experience. *J Korean Med Sci* 2016;31:1397-1402.
4. Morin DP, Iwai S. Iatrogenic Twiddler's syndrome. *J Interv Card Electrophysiol* 2010;29:135-137.
5. Arindam P, Achyut S, Imran A, Naveen Ganiga Sanjeeva C, Rabin C. Pacemaker Twiddler's syndrome: review through a case report. *J Cardiovasc Dis Res* 2015; 6:148-151.
6. Morales JL, Nava S, Marquez MF, et al. Idiopathic lead migration: concept and variants of an uncommon cause of cardiac implantable electronic device dysfunction. *JACC Clin Electrophysiol* 2017;3:1321-1329.
7. Wang Y, Hou W, Zhou C, et al. Meta-analysis of the incidence of lead dislodgement with conventional and leadless pacemaker systems. *Pacing Clin Electrophysiol* 2018;41:1365-1371.
8. Schinkel AF, Vriesendorp PA, Sijbrands EJ, Jordaens LJ, ten Cate FJ, Michels M. Outcome and complications after implantable cardioverter defibrillator therapy in hypertrophic cardiomyopathy: systematic review and meta-analysis. *Circ Heart Fail* 2012;5:552-559.
9. Osoro M, Lorson W, Hirsh JB, Mahlow WJ. Use of an antimicrobial pouch/envelope in the treatment of Twiddler's syndrome. *Pacing Clin Electrophysiol* 2018;41:136-142.
10. Sobstyl MR, Zabek M, Brzuszkiewicz-Kuzmicka G, Pasterski T. Dual anchor internal pulse generator technique may lower risk of twiddler's syndrome: a case series and literature review. *Neuromodulation* 2017;20:606-612.
11. Shandling AH, Ellestad MH, Castellanet MJ, Messenger JC. Dacron-woven pacemaker pouch. Influence on long-term pacemaker mobility. *Chest* 1991; 99:660-662.

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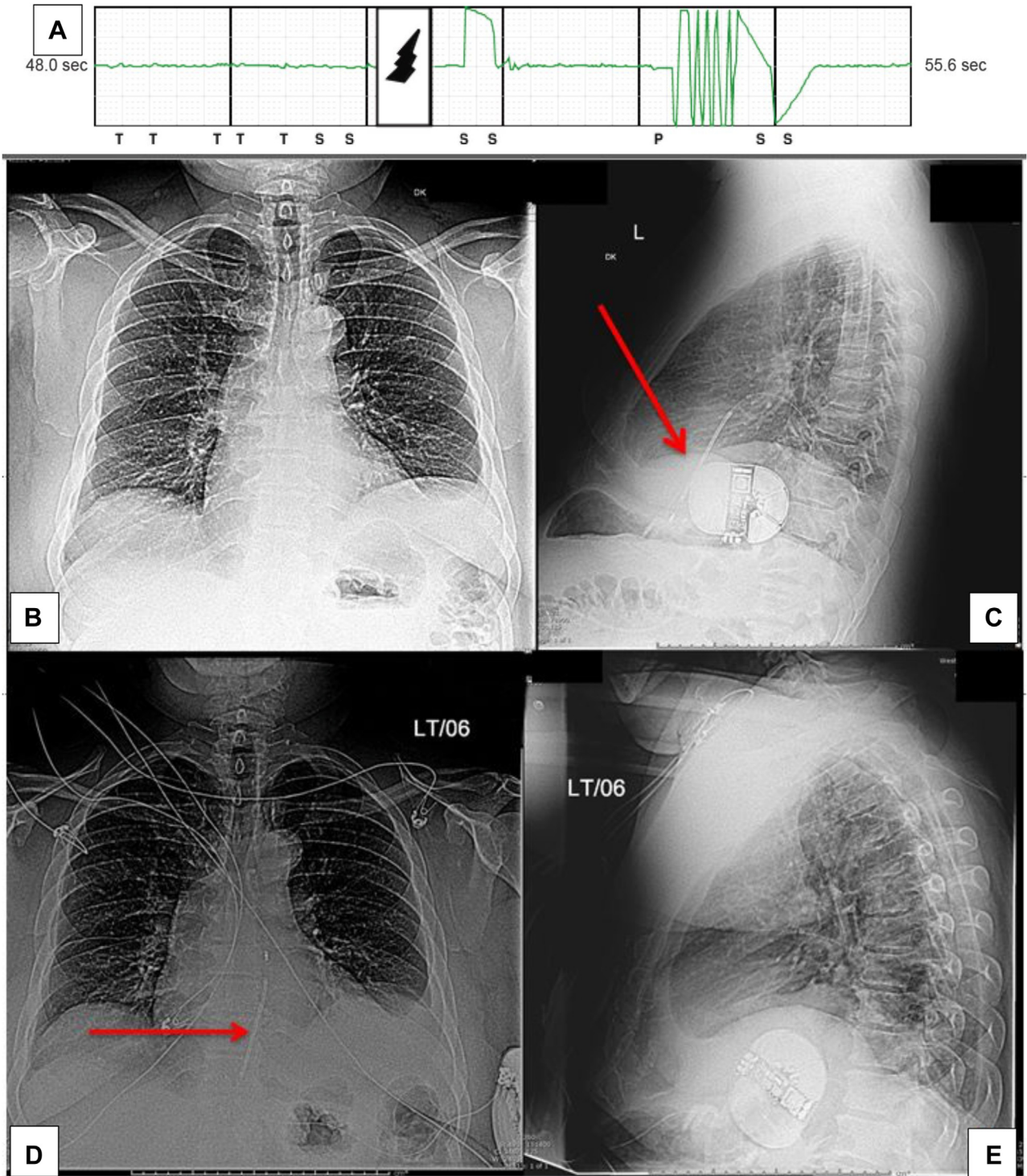


Figure 1 A: Electrogram showing inappropriate shock. B, C: Anterior-posterior/lateral chest radiographs showing lead dislodgement. D, E: Anterior-posterior/lateral chest radiographs after initial implantation. Red arrows indicate lead tip. Note difference of pulse generator orientation in pocket compared to original implant.