Internal Twiddler’s Syndrome: Not Your Average Knot

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Case presentation
An 83-year-old female with a dual-chamber pacemaker, chronic kidney disease, and coronary artery disease status post coronary artery bypass grafting developed severe symptomatic aortic stenosis and underwent bioprosthetic aortic valve replacement. Her postoperative course was complicated by sustained monomorphic ventricular tachycardia.

A single-chamber ventricular pacemaker (Guidant (St. Paul, MN)/CPI 1176 Meridian SR generator; Guidant/ CPI 4261 right ventricular (RV) lead) was originally implanted 8 years ago with a right sided pre-pectoral pocket. Her device was upgraded to a dual-chamber pacemaker (Guidant/CPI Insignia Entra generator; Guidant/CPI 4135 Dextrus atrial lead) 1 year ago due to the development of pacemaker syndrome. The RV lead was left intact and not resutured. There was no evidence of twiddling at that time. Interrogation of the device prior to her aortic valve surgery showed an increase in ventricular capture threshold (2.5 V at 0.4 ms from 1.5 V at 0.4 ms), a decrease in ventricular sensing (2.6 mV from 6.7 mV), and an increase in ventricular impedance (1490 Ω from 1150 Ω) compared with 1 year ago. Threshold, sensing, and impedance of the atrial lead were stable and within acceptable limits.

Owing to sustained ventricular tachycardia, she underwent upgrade to an implantable cardioverter-defibrillator (ICD) for secondary prevention of sudden cardiac death. Her pre-procedural chest X-ray revealed a knot involving her RV pace-sense lead and entangled leads in the device pocket consistent with twiddling (Figure 1). These findings were confirmed with fluoroscopy during her procedure (Figure 2). Her central venous system was patent, the RV pace-sense lead was capped, and a new single coil defibrillator lead (CPI/Guidant 0180 Reliance SG) and ICD generator (Boston Scientific (St. Paul, MN) E110 Teligen) were implanted without complication.

Discussion
Twiddler’s syndrome is a well-known complication of implanted cardiac devices. Patients manipulate the device within the pocket often leading to lead malfunction and even dislodgement. It has been reported in both pacemakers and ICDs, typically resulting in coiling of the leads within the generator pocket. Our patient developed dysfunction of her RV lead, but unlike previous reports there was no previous knotting of the RV lead within the heart in addition to the device pocket. Interestingly, her atrial lead was unaffected. There are no technical features of the RV lead (Guidant/CPI 4261) to explain this difference, and it is likely due to differential tie-down techniques as the pace-sense RV lead was tied down with absorbable suture and the atrial lead was tied down with non-absorbable suture. The use of absorbable suture on the RV lead appears to have allowed transmission of torque to the distal lead body and coiling within the heart. To our knowledge this is the first reported case of “internal twiddling.”

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Figure 1: Pre-procedural lateral chest X-ray demonstrating twisting and knotting of the right ventricular pace-sense lead within the right ventricular cavity.
References


Figure 2: Fluoroscopic image obtained at the time of device upgrade confirming the knot in the right ventricular lead.