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A false sense of security: Twiddler syndrome presenting ten days after routine interrogation

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Introduction: Twiddler syndrome is a rare but potentially serious mechanical complication in patients with cardiac implantable electronic devices (CIEDs). It results from rotation of the pulse generator around its longitudinal axis within the subcutaneous pocket.[1,2] This may cause the leads to twist around their own axes or become entangled, potentially resulting in lead retraction, dislodgement, or damage and consequent device malfunction. Predisposing factors include inadequate lead fixation, an oversized subcutaneous pocket, obesity, female sex, and conscious or unconscious manipulation of the device by the patient.

Case report: 69-year-old woman with obesity, persistent atrial fibrillation (AF), and a rapid ventricular response despite medical therapy underwent implantation of a VVIR pacemaker in September 2017, with an active-fixation lead positioned on the right ventricular septum, followed by radiofrequency ablation of the atrioventricular node. Device function was normal at follow-up 10 days later. She subsequently developed progressive dyspnea and reduced exercise tolerance. Three weeks after implantation, she was admitted with decompensated heart failure and treated with intravenous furosemide. The ECG showed AF with a regular narrow-QRS escape rhythm at 56 beats/min, below the programmed pacing rate of 70 beats/min, with pacing spikes but no ventricular capture. The radiologist reported a lead that could be "followed into the right ventricle". Fluoroscopy confirmed lead retraction and a generator-pocket appearance typical of Twiddler syndrome. The lead was repositioned at the right ventricular apex, and the generator was secured to the muscle. The patient confirmed that she had manipulated the device. During nine years of follow-up, including an upgrade to CRT-P in 2021 because of pacing-induced cardiomyopathy, and generator replacement in 2026, no recurrence was observed.

Conclusion: Prevention of Twiddler syndrome includes adequate fixation of the lead, careful creation of the device pocket, and patient education regarding avoidance of device manipulation. Treatment is based on surgical revision, repositioning and secure fixation of the generator, and assessment of lead integrity and function, with lead repositioning or replacement as required. In selected patients, leadless pacemakers may represent an alternative to conventional systems.

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