



## Pectoral muscular twitching: a rare manifestation of spontaneous twiddler syndrome

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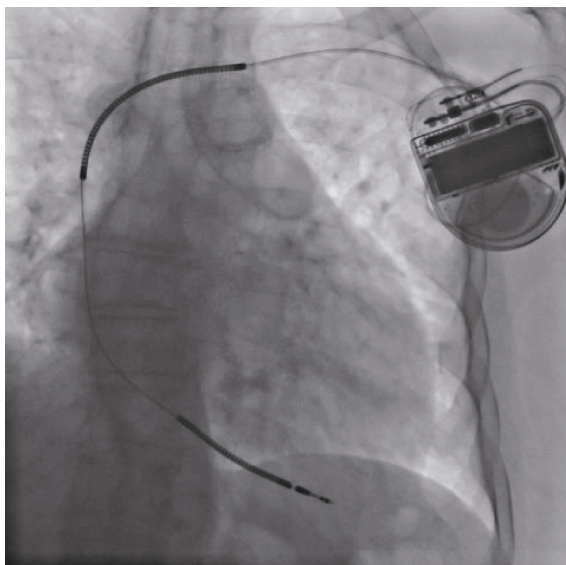
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The twiddler syndrome is an uncommon cause of pacemaker malfunction, in which twisting or rotating of the device in its pocket results in lead retraction or coiling and subsequent malfunction of an implanted devices such as a pacemaker, an implantable cardioverter-defibrillator (ICD), cardiac resynchronization therapy (CRT).<sup>[1,2]</sup>

This report describes a rare case of twiddler syndrome after implantation of a ICD that manifested with pectoral muscular twitching.

A seventy-three years old woman who underwent primary prevention single-chamber ICD implantation (Figure 1) due to ischemic cardiomyopathy, presented to us with feeling of stinging and twitching around the generator in left

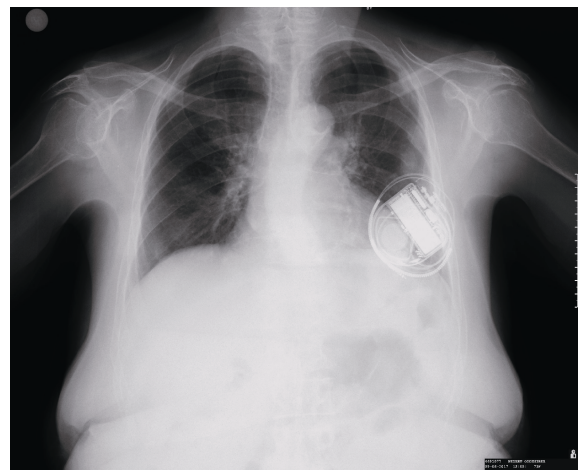


**Figure 1.** Fluoroscopy view taken just after implantation showing normal position of the pulse-generator and normal appearing lead without any twirling.

pectoral region. She had no any other complaints at presentation. Chest radiograph showed that the right ventricular (RV) lead had been retracted back to the left pectoral region and encircling the generator (Figure 2). She had no cognitive deficit and denied manipulating the pacemaker generator.

ICD (Medtronic, Maximo II VR D284VRC) was implanted seven weeks ago, from the left side in the usual way. RV lead (Durata™ defibrillation lead, St Jude Med) was active fixation. At implant, adequate sensing, pacing and impedance values were confirmed (R wave 8.7 mV, threshold 0.9 V at 0.5 ms, impedance 472 Ohm). ICD interrogation performed on last admission of the patient showed a decrease of RV wave amplitude (0.3 mV). RV pacing, RV and SVC defib impedance were 285, 3, 11 Ohm, respectively.

RV lead was extracted and replaced by new one under fluoroscopic guidance with satisfactory parameters (Figure 2).



**Figure 2.** Chest radiograph showing the coiling of the right ventricular lead around the pulse generator of an implantable cardioverter defibrillator.

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The lead was secured with non-absorbable double sutures around the sleeves and generator was to the pectoralis muscle. Since then, the patient is being followed up with no recurrence of any complication or malfunction.

The twiddler syndrome is caused by the spontaneous or self-induced manipulation of the pacemaker pulse generator around its central axis within the pocket, resulting in retraction and dislocation of the leads. It has a reported frequency of around 0.07%–7%, more commonly reported among elderly females and obese patients.<sup>[3–5]</sup> The majority of cases are diagnosed within the first year of implant,<sup>[6,7]</sup> although it can occur at any time after device implantation. Our old female patient presented after seven weeks of ICD implantation.

Manifestations of twiddler syndrome are depending on the final site of the dislodged lead. In most cases, the keys to confirming twiddler syndrome have been loss of capture, stimulation of noncardiac structures or inappropriate ICD discharge. Leads that get dislodged further up may stimulate the phrenic nerves causing diaphragmatic contractions. Further coiling and withdrawal of the lead, leads to stimulation of the brachial plexus, resulting in rhythmic arm twitching.<sup>[1,3,7]</sup> Recently, Sekimoto *et al* reported a rare case of twiddler syndrome, which was detected as an alert for intrathoracic impedance as an indicator of heart failure. The recovery of thoracic impedance after relocation in this case make the possibility that the intrathoracic impedance might have been directly affected by the length between the lead and the generator.<sup>[8]</sup> Our patient presented with a twitching in left pectoral region due to pectoral muscle stimulation by the retracted lead encircled the pulse generator.

Twiddler syndrome is more commonly recognized in obese patients due to the increased subcutaneous tissue, possibly providing less support from surrounding tissue to the pocket. An associated increased laxity of the subcutaneous tissues, particularly in elderly patients facilitates further dislodgement of device. Additionally, a smaller sized implanted device relative to its pocket permit their rotation within the skin pocket.<sup>[4,6,7]</sup> Another factor is postimplant pocket haematoma formation. This may cause autocleavage of the original dissection plane. Once reabsorbed, the hematoma of the residual “space” allows for greater displacement of the device.<sup>[9]</sup>

Goldenberg, *et al.*<sup>[10]</sup> reported a case of twiddler syndrome as a consequence of device rotation without manipulation of the pocket, thus a spontaneous “Twiddler’s” phenomenon. As in our case, most patients deny manipulating their device.<sup>[4,7,9]</sup> There have also been reports of cases in which patients admit device manipulation, so patients should be educated in this subject.<sup>[11]</sup> It has recently been reported in a case series that some device models may also

cause spontaneous rotation due to their shape and thus this undesirable phenomenon.<sup>[9]</sup>

The chest X-ray is the simplest and most vital diagnostic tool to diagnose twiddler syndrome,<sup>[7]</sup> as it is rapid and gives a clear image of the lead coiling and device rotation. Treatment of diagnosed cases include repositioning of the dislodged leads, implantation of a new lead and repositioning of the pulse generator. Avoiding pocket-CID mismatch, correct fixation of the leads and the device with a non-absorbable suture to the pectoralis fascia and instructing patients not to manipulate their device may prevent further occurrences of twiddler syndrome.<sup>[9]</sup>

Although twiddler syndrome is certainly uncommon, it remains a significant clinical problem in ICD patients, as it will lead to failure to detect or treat potentially lethal ventricular arrhythmias. Fixating the device and the leads at the pectoral fascia and proper patient education can prevent this complication.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

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