Case Report

Twiddler's syndrome in a patient with CRT-D device – A case report

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ABSTRACT

Twiddler's syndrome is a rare cause of pacemaker lead dislodgement. A 61-year-old female patient was implanted with a defibrillator capable of cardiac re-synchronization therapy (CRT-D); 10 months later, she presented with uneasiness and vibratory sensations in the chest. Fluoroscopy revealed rotation of the pulse generator along its longitudinal axis and dislodgement of all three leads. Diagnosis of Twiddler's syndrome was made.

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1. Introduction

Twiddler's syndrome is a rare cause of pacemaker lead dislodgement. It is characterized by deliberate or spontaneous rotation of the pulse generator leading to lead malfunction. It was first described by Bayliss in 1968. In this case report, we describe dislodgement of all three leads of CRT-D due to Twiddler's syndrome.

2. Case presentation

A 61-year-old female (dilated cardiomyopathy with severely depressed left ventricular ejection fraction, left bundle branch block and New York heart failure class III symptoms, diabetes, hypothyroidism) was implanted with a defibrillator capable of cardiac re-synchronization therapy (CRT-D St Jude unify) in August 2014. For the right atrium and the right ventricle active screw-in leads were used. The left ventricular lead was a passive fixation bipolar lead that had been implanted in a postero-lateral vein.

At the time of the last regular in-office follow-up (March 2015), the patient's heart failure symptoms had improved and ICD interrogation revealed no remarkable findings.

In June 2015, she presented with complaints of uneasiness and vibratory sensation over the chest and epigastric area for 3 weeks. CRT-D interrogation showed loss of function of all three leads. She was taken up for fluoroscopy which showed rotation of the pulse generator along its longitudinal axis with complete dislodgement of all three leads (Figs. 1 and 2). The right atrial lead was retracted with its tip into the superior vena cava, the right ventricular lead into the right atrium, and the left ventricular lead to the coronary sinus ostium. The patient’s daughter had noticed that her mother often scratched at the device implant site. The patient herself

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denied any manipulation to the device. She was advised for re-implantation of the CRT-D device.

3. Discussion

Twiddler’s syndrome is a rare cause of macro-dislodgement and other lead failures (e.g. insulation brakeage and lead fracture). Elderly and obese patients appear to be at increased risk because the presence of loose subcutaneous tissue allows for rotation of the pulse generator in its pocket.\(^7\) Possible complications due to Twiddler’s syndrome are syncope in cases with pacemakers and inappropriate shock, lack of appropriate shock, and loss of pacing in ICDs and CRT-Ds. Phrenic nerve stimulation by the dislodged leads can cause diaphragmatic contractions, and the stimulation of the brachial plexus can cause twitching of arm.\(^3\) Another entity, which is associated with pulse generator rotation, is Reel syndrome.\(^6\) The basic difference between Twiddler’s and Reel being that in the former, the device rotates along its longitudinal axis, and in the latter it rotates in transverse axis. In this case, the pulse generator has clearly rotated along its longitudinal axis, which is typical of Twiddler’s syndrome.

To the best of our knowledge, there is no previous report of dislodgement of all three leads of CRT-D device due to Twiddler’s syndrome.

The use of screw in leads as well as subpectoral implantation has been described as possible preventive strategies, but these are not always successful;\(^5\) in our case, the patient had screw in leads in the right atrium and ventricle.

Conflicts of interest

The authors have none to declare.

References