

Case Report

Lead Dislodgement Related to the Unique Design of an Implanted Cardioverter Defibrillator during the Late Phase after Implantation: A Case of Twiddler Syndrome

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Keywords

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Abstract

Twiddler and Reel syndromes are rare complications causing lead dislodgement. Twiddler syndrome, whereby twisting or rotating of the device over the long axis in its pockets, results in lead dislodgement and device malfunction. Previous reports showed unique device design easily caused Twiddler syndrome and lead dislodgement. The majority of lead dislodgement occurred in the perioperative time. However, we present a rare case a unique device design that caused Twiddler syndrome and resulted in lead displacement during the late phase after implantation.

ABBREVIATIONS

ICD: Implanted Cardioverter Defibrillator

INTRODUCTION

Twiddler syndrome is caused by the spontaneous or self-induced manipulation of the generator over the axis in the pocket. Previous reports showed the frequency of twiddler syndrome was around 1% [1], and longitudinal symmetric nature of the device design has a higher risk of intrapocket device rotation based on its unique shape [2]. A lead dislodgement during the late phase, is a rare complication. The majority of a lead dislodgement occurred in the perioperative time. We present the case of the atrial lead dislodgement associated with Twiddler syndrome caused by the unique device design at 13 months after implantation.

CASE PRESENTATION

A 78-year-old man with ischemic cardiomyopathy underwent coronary artery bypass and received a dual-chamber implanted cardioverter defibrillator due to ventricular tachycardia. We

performed an extrathoracic puncture and inserted two leads in the pocket. An active fixation lead (Tendril STS model 2088TC-46; St. Jude Medical, Minneapolis, MN, USA) was placed in the right atrial appendage. The stimulation threshold was 1.3 V at 0.4 ms, and the atrial wave amplitude was 2.0 mV. The active fixation ventricular lead (Durata model 7122Q-58; St. Jude Medical) was positioned at the right ventricular apex. The stimulation threshold was 0.6 V at 0.4 ms, and the R-wave amplitude was 12.5 mV. The suture sleeves were secured with one silk (size: 1-0) suture for each lead. We performed a tug test on those leads and ensured no movement of both leads. We implanted a de novo pulse generator (Fortify Assura; St. Jude Medical) that was stitched to the left pectoral muscle and underlying fascia. The implanted cardioverter defibrillator pocket was appropriately matched with the device size, and the implanted cardioverter defibrillator was anchored to the underlying muscle with silk (size: 1-0) for stability (Figure 1a). At one and three month follow-up, chest radiography indicated that the position of the leads and generator had not changed compared with the original implantation site. When the patient attended our hospital for routine follow-up at

9 months after implantation, chest radiography revealed that the header of the implanted cardioverter defibrillator had dropped approximately 2.5 cm and turned 180 degrees over the long axis compared to the original implantation location. The atrial lead was twisted in the pocket and pulled back toward the head. However, the ventricular lead was still on the right ventricular septum, as it was during implantation (Figure 1b). The atrial lead threshold was increased to 1.7 V at 0.4 ms, and the ventricular lead was not changed. Both leads were not retracted out of their respective implantation sites. Therefore, we decided to perform follow-up observation according to the routine schedule and instructed him not to manipulate his device pocket again. At 13 months after implantation, the atrial lead threshold was more than 7.5 V at 0.4 ms, and chest radiography showed dislodgement of the atrial lead (Figure 1c) without any change in the position of the pulse generator. However, the position of the twisting lead and sleeve had changed compared with 9 months follow-up period. The atrial lead moved inside the shock lead in the pocket, and rotation of the generator caused curvature of the atrial lead. The atrial lead did not interfere with the shock lead and documented atrial arrhythmia. And he did not need atrial and ventricular pacing. Therefore, we changed the pacemaker mode from DDD to VVI (40bpm). No device malfunction was observed by remote monitoring.

DISCUSSION

Lead dislodgement is a significant complication of device implantation. The incidence of lead-related re-intervention was 4.4% for patients who received a cardiac rhythm device [3]. The majority of lead dislodgement occurred in the perioperative time [3]. However, lead dislodgement during the late phase after implantation is a rare complication. Our patient experienced

atrial lead retraction during the late phase because of Twiddler syndrome. Twiddler syndrome is a commonly recognized cause of device malfunction due to the rotation of the device in the pocket on its long axis [4]. Twiddler syndrome has been reported to have a frequency of approximately 1% and especially 7% in elderly patient.¹ Its risk factors include female sex, large implant pocket, obesity, young age (children), old age (elderly patients), hematoma, and the spontaneous or self-induced manipulation of the generator [2]. Bozyel S, et al. presented that increased laxity of the subcutaneous tissues in elderly patients facilitates dislodgement of device.¹ Our patient was an old man and had excess subcutaneous laxity. The fixation of the leads and the device under the greater pectoral muscle might prevent dislocation. However, the subcutaneous device pocket is easy to make and less hemorrhage. 9 months after implantation, the generator turned 180 degrees over the long axis. The atrial lead was twisted in the pocket and pulled back toward the head. However, atrial and ventricular leads did not cause dislodgement. On careful history taking, he denied manipulation of his device pocket. However, there was a possibility that our patient rotate the device unconsciously. For 13 months, only an atrial lead had retracted out of the original implantation site. However, a ventricular lead and generator had not changed positions compared with 9 months follow-up period. Chest radiography revealed increased atrial lead curvature in the pocket compared with 9 months after implantation, and the atrial lead moved inside the shock lead in the pocket (Figure 1). Rotation of the generator caused curvature of the atrial lead. We speculated that loosening of the suture only on the atrial lead fixation sleeve caused atrial lead dislodgement due to rotation of the generator. Because the ventricular shock lead is strongly fixed behind the pocket, Twiddler syndrome could not cause ventricular lead dislodgement. In addition, several

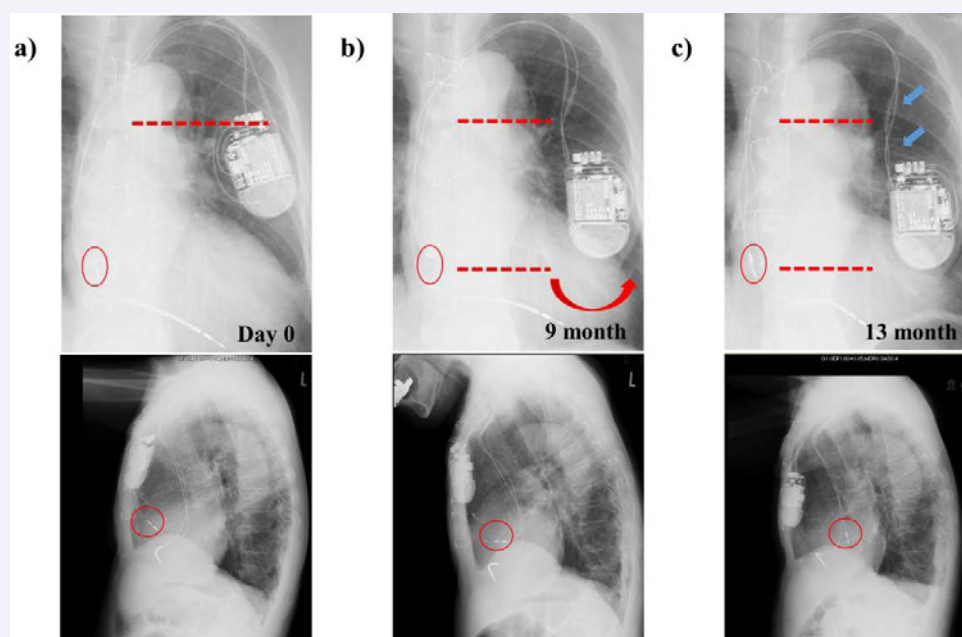


Figure 1 Chest radiographs showing the transition associated with leads and a generator in a time series

a) Original implantation sites of the leads and generator. b) At 9 months after implantation, the generator dropped approximately 2.5cm compared to the original implantation location and turned 180 degrees over the long axis. The atrial lead was twisted in a pocket and pulled back toward the head. c) At 13 months after implantation, the atrial lead curvature was enlarged (arrow), and dislodged. The ventricular lead did not change the site.

reports showed that the unique design of the device easily caused Twiddler syndrome [2,5]. Gul et al presented three case reports of Twiddler syndrome. They concluded that the longitudinal symmetric nature of the implanted cardioverter defibrillator design leads to a higher risk of intrapocket device rotation. Only one anchored part of the longitudinal device was susceptible to spontaneous torsion [2]. In our patient, ligation of the atrial suture sleeve was insufficient, the subcutaneous tissue was very loose, and the longitudinal device was implanted. We believe those factors caused Twiddler syndrome during the late phase after implantation.

CONCLUSION

Twiddler syndrome is a rare complication. However, unique device formation easily causes lead dislodgement. Making the device pocket under the greater pectoral muscle is the one of solutions. Additionally device pocket with an appropriate size, suturing the sleeve tightly, and anchoring the generator to the pectoral muscle strongly should prevent twisting of the device for elderly patients.

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