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Delayed presentation of pacemaker Reel Syndrome and lead damage: Coincidence or consequence?

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ABSTRACT

Reel syndrome is a pacemaker lead early dislodgment, characterized by reeling-in of the lead(s) without being damaged. We herein present a case of an 86-year-old woman, with medical history of single chamber pacemaker implantation two years ago, admitted in cardiology department with complete AV block. Chest-Xray revealed ventricular lead coiling around and behind the pacemaker device. Urgent extraction of the previous pacemaker was performed; however, the lead damage made its repositioning unfeasible. Successful implantation of single-chamber pacemaker has been made. This clinical case highlights the importance of adequate follow-up to timely identify lead dislodgement, avoid lethal complications and lead fracture.

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1. Introduction

Pacemaker-twiddler's syndrome is a rare but potentially lethal complication of cardiac pacemaker treatment. First described by Bayliss and al. in 1968 [1], "Reel syndrome" has been described as a variant of Twiddler syndrome, characterized by reeling-in of the pacemaker lead(s) [2]. Presenting symptoms are often related to loss of capture leading to dyspnea, dizziness and syncope. Thus, urgent management is mandatory due to sudden death risk. Herein, we report the case of an 86-year-old woman with Reel syndrome whose condition has recently been revealed two years after pacemaker implantation by a syncopal attack. In our case, both late lead dislodgment and fracture were described which is uncommon in Reel Syndrome.

2. Case report

An 86-year-old woman presented to the emergency room with a femoral neck fracture that occurred right after a syncopal attack.

Her medical history revealed single chamber pacemaker implantation in 2019 owing to complete AV bloc. However, she was

lost to cardiac follow up.

She also had dementia and the family reported involuntary rhythmic right arm movements several months ago.

Upon arrival at the emergency room, she had a pulse of 36/min, and a complete atrioventricular block on the electrocardiogram (ECG) with pacemaker spikes and failure to capture (Fig. 1). Chest X-ray urgently performed revealed ventricular lead coiled around and behind the generator secondary to rotation of the pulse generator on its transverse axis (Fig. 2).

The patient was admitted immediately to the cardiology department and underwent surgery for adjustment of the device. The previous permanent pacemaker had been extracted from its submuscular pocket along with the lead coiled around it (Fig. 3- A & B). The lead's obvious damage (Fig. 3- C) made its repositioning unfeasible.

Successful implantation of a single chamber pacemaker has been made on the same side (left side) and the pulse generator was tied with a nonabsorbable suture to the pectoral muscle.

Further recovery was uneventful, and the patient was transferred to the traumatology department in stable condition.

Since replacement of the lead, no subsequent lead problems have been detected, and the pacemaker system was functioning normally at both her three and six months follow-up visits.

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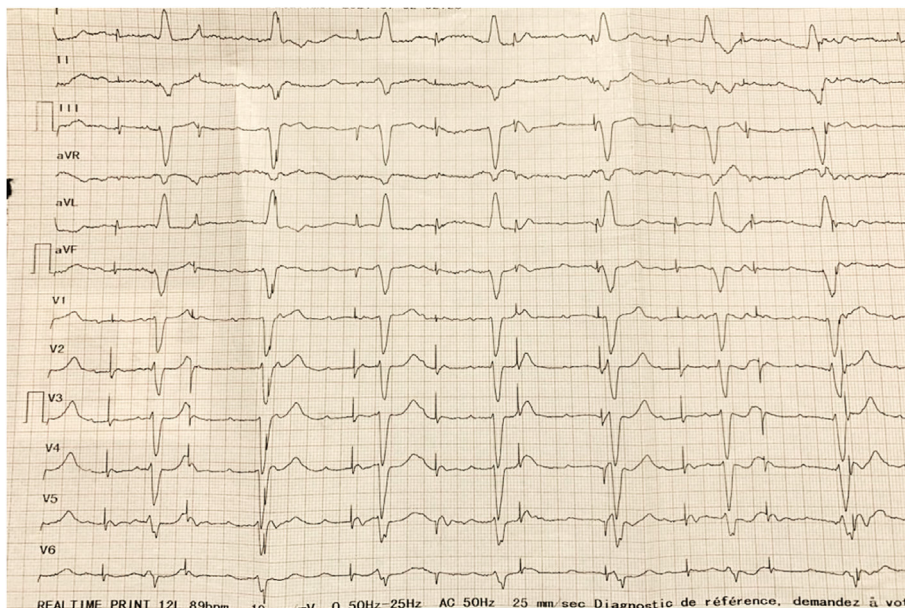


Fig. 1. Electrocardiogram on admission showing complete heart block with inadequate capture.



Fig. 2. Chest X-ray demonstrating ventricular lead coiling around and behind the pacemaker device.

3. Discussion

Dislodgement of pacing lead(s) is a well-known complication of device implantation. First described by Bayliss and al. in 1968, Twiddler syndrome [1] was identified as a dysfunction of a cardiac implantable electronic device lead resulting from direct manipulation or spontaneous rotation of the generator.

Various mechanisms of lead macro-dislodgement leading to device malfunction have been described: Twiddler's syndrome is the rotation of the generator on its long axis, which induces damage to the leads; if it's a dual lead, the x-ray will show tangling of the leads. In contrast, there is a rotation of the generator on its

transverse axis in Reel syndrome but no damage to the leads. The leads will be coiled around the generator and retracted. Finally, Ratchet syndrome is attributed to retraction and electrode dislocation with ratcheting but without coiling of the generator due to progressive retracting of the electrodes from their fixing protections [3].

Reel syndrome is commonly observed within months of the placement of the generator compared to Twiddler which takes years to occur [4]. In our case, Reel syndrome occurred two years after device implantation, which is uncommon.

Twiddler and Reel's syndromes share common etiology: female gender, obesity, large pocket, advanced age, dementia, and deep brain stimulation can be listed as contributing factors [2,5]. Our patient was a female in the geriatric population, with both obesity and dementia.

Other risk factors include participating in sports requiring wide and repetitive movements of the arms, such as golfing and swimming [6].

Until lead dysfunction occurs, Reel Syndrome remains silent and acute complications are either attributed to loss of capture leading to dyspnea, dizziness, exercise intolerance and syncope or to extra-cardiac stimulation inducing symptoms such as dysphonia, hiccups, thoracic muscle or brachial plexus stimulation ... [7].

In our patient, the diagnosis was made throughout a syncopal attack leading to femoral neck fracture.

Chest X-ray is a convenient and simple diagnosis tool, with the possibility to distinguish between these three syndromes.

Lead dysfunction usually presents during the first weeks in Twiddler syndrome, but have been described as early as 17 hours and as late as one year after device implant [6,8]. Interestingly, in Reel syndrome not all leads are affected equally, and «selective» cases, have been described [9].

Our patient only had single chambre pacemaker, with apparent lead damage in our operative finding (Fig. 3), we assume that delayed diagnosis is the main cause of this condition. A thorough search of the relevant literature yielded only one related article describing lead damage in Reel syndrome [9].

To prevent device rotation, several techniques have been proposed including the creation of a small pocket; sub pectoral implant

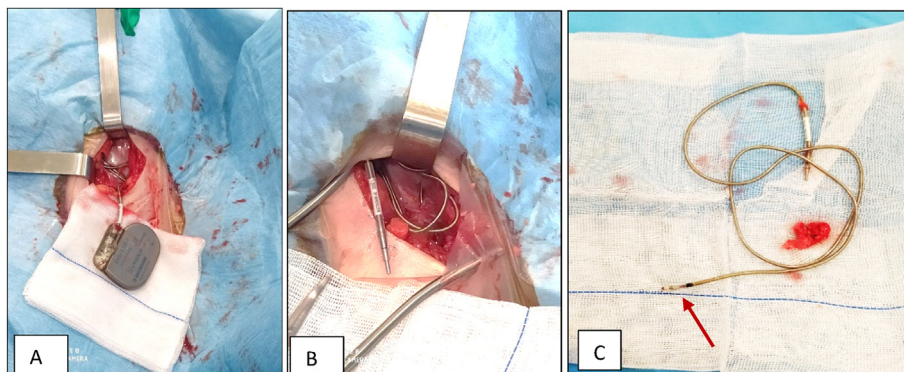


Fig. 3. A and B = operative views demonstrating Reel Syndrome with lead twisted behind the pacemaker. C = Lead damage (red arrow).

of the device; use of non-absorbable suture for fixation or polyester cases; use of active fixation leads and even immobilization of the upper extremity in the first week after implantation. Unfortunately, all these techniques seem doomed to failure. Recurrences have been reported with all of these. However, using leadless pacemaker implantation will also likely reduce lead dislodgement and hence, prevent Twiddler syndrome from occurring [10].

In conclusion, pacemaker lead dislodgement is an unusual occurrence. It is a clinically serious and potentially life-threatening condition due to sudden death risk. Early dislodgments are more frequent than late. This clinical case highlights the importance of adequate follow up to timely identify lead dislodgement and avoid lethal complications beside lead fracture.

4. Key teaching points

- Lead dislodgement is a clinically serious and potentially dangerous event.
- Reel Syndrome typically occurs within months after pacemaker implantation, but our patient presented two years after.
- Lead fracture can be explained both by the trauma due to the episode of syncope and to patient several manipulation.
- This case report highlights the importance of regular follow-up to timely identify lead dislodgement and avoid lethal complications beside lead fracture.

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Authors' contributions

All authors have read and approved the manuscript.

S.H. is the main author who managed the patient. F.C and F.I. are the co-authors who analyzed the patient data and were major contributors in writing the manuscript. I.F and M.C supervised the management of the patient and revised the manuscript.

Consent for publication

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

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