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Rapid firing Double twiddle trouble, a new variant of twiddler syndrome

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1. Case report

A 65-year-old woman with permanent atrial fibrillation was admitted with diastolic heart failure (HF) secondary to rapid ventricular response. She had undergone single chamber pacemaker implantation (Boston Scientific Ingenio SR) two years earlier for symptomatic bradycardia (Fig. 1A). During the current admission, a chest radiograph and fluoroscopy revealed that the lead (Guidant[®] FINELINE[™] Model 4457, passive fixation) had become dramatically twisted at 2 sites: in the pacemaker pocket and within the right atrium (Fig. 2A, B). Nevertheless, pacing/ sensing and impedance parameters remained normal. In retrospect, a radiograph performed 2-months post-implant due to a respiratory tract infection already showed rotation of the impulse generator and the development of two initial loops in the lead occurring at these 2 sites simultaneously (Fig. 1B). However, this was unnoticed at that time.

An echocardiogram was performed in the hospital showing a left ventricle ejection fraction of 50% with no wall motion abnormalities or significant valve disease. The patient was initially managed with optimization of HF medications. Due to rate control being unachieved and recurrent episodes of HF, atrioventricular node ablation was performed, and the patient was offered an upgrade to a biventricular pacemaker with a new right ventricular lead [1].

ABSTRACT

The twiddler syndrome results in retraction and coiling of the lead in the pacemaker pocket with subsequent pacemaker malfunction. If the lead's parameters are not affected, then the problem can be corrected by lead repositioning. We describe the first reported case of a dramatic pacemaker lead coiling with a double level of lead compromise, with one of them being intra-cardiac. This condition affected our treatment strategy, as the repositioning of the lead was unfeasible.

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2. Discussion

Lead dislodgement is a relatively common complication of pacemaker implantation [2]. Different mechanisms of lead macrodislodgement leading to device malfunction have been described. The twiddler syndrome is caused by the spontaneous or selfinduced twisting of the generator over the axis defined by the lead, causing the typical twisted lead image [3]. In the "reel syndrome", the generator rotates over its transverse axis resulting in a lead coiling around the generator [4]. The "ratchet syndrome" occurs due to progressive lead displacement through its fixation parts without generator rotation over any of its axes [5].

Although many variants of twiddler syndrome have been reported, none describes such dramatic twisting with a double level of lead compromise, with one of them being intra-cardiac.

The reason for the intra-atrial coiling remains unclear. A hypothesis would be that the stitch to the lead sleeve was not tight enough and a large amount of repetitive torque was transmitted to the lead. As the patient denied manipulation of the device, it may have occurred as a result of movements of the ipsilateral arm or inadvertent manipulation of the pulse generator. Although this mechanism is relatively common, an intra-atrial coiling has not been seen before, as the lead is usually retracted with subsequent coiling in the pocket. In our case, the lead was also retracted, but the coiling took place in the pocket and right atrium. Interestingly, the radiograph performed 2-months post-implant suggests that the coiling occurred at the same time in both sites.

Consideration of a new lead was the result of concerns about the long-term reliability of this lead. Although the lead integrity was not compromised, our patient had two potential sites of lead damage, which may arise from continuous shear stress or torque

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Fig. 1. Old chest radiographs showing the lead in situ (A, post-procedure), and the rotation of impulse generator with the development of two loops in the lead (B, 2 months post-procedure).



Fig. 2. (A) Current chest radiograph showing the lead retracted and coiled at the pacemaker pocket and within the right atrium. (B) Fluoroscopy in the right anterior oblique projection performed prior to the atrioventricular node ablation shows the intra-atrial coiling of the lead in detail.

from multiple coiling sites along the length of the lead. In contrast to other cases, where the problem was corrected by lead repositioning rather than replacement, the intra-atrial coiling did not allow us to reposition it. Should a lead extraction be required in the future, consideration should be given for a combined subclavian and femoral approach.

3. Conclusion

We describe a new variant of twiddler syndrome with retraction of the lead resulting in twisting at two different levels. This altered our treatment strategy, as lead repositioning was unfeasible. Minimizing pocket size and correct fixation of the lead and device to the pectoralis fascia with appropriate patient education may prevent this complication.

Conflict of interest

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Author contribution

Contributions for drafting article (A.G.C. and P.S.G.H), figure editing (A.G.C), and critical revision (A.G.C., J.S.H and G. A.).

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