



Very-late onset twiddler syndrome as an unusual cause of syncope

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Received: 16 July 2019 / Accepted: 13 August 2019
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A 59-year-old female was implanted with an ICD in 2011. The patient had a subpectoral implantation followed by an unremarkable course. In January 2018, battery replacement was performed and an Ilivia 7 DR-T (Biotronik, Germany; DDDR 60-140/min) was implanted, the leads remained untouched. In August 2018, a remote monitoring alert was triggered due to intermittent loss of right ventricular capture mimicking short ventricular intervals. In-house checks showed a stable pacing threshold of 1.7 V/0.4 ms and an impedance of 430 Ω . A fixed higher pacing output was programmed. Subsequently, we observed a continuous increase in the pacing threshold in the remote monitoring (Fig. 1). We interpreted this as a changing electrode-tissue-interface and adjusted the pacing output. In April 2019, a sudden drop in the R-wave amplitude was registered and a few days later, the patient experienced syncope. In the emergency unit, she was in sinus rhythm, but no capture was obtained with maximum stimulation output, indicating ventricular exit block as the reason for syncope. The chest radiograph showed a lead dislodgment and operative revision was performed (Fig. 2). The patient denied device manipulations, traumas, or weight changes.

This severe very-late onset twiddler syndrome many years after lead implantation with “self-extracted” tangled leads is unusual. The ingrowth of leads and the generator after implantation decrease the susceptibility to idiopathic lead migration in the absence of risk factors [1]. The most likely mechanism of lead migration in this case was ratcheting of the leads caused by torque induced by the pectoral muscle. Remote monitoring revealed equivocal electrode parameter changes several months prior to the syncope. However, the changes cannot be considered as typical failure signs of this ventricular lead [2]. Signs for lead dislodgment typically encompass a drop in sensing amplitude and impedance, with a rise of the pacing threshold [3]. This case illustrates that an isolated progressive increase of the pacing threshold can be the only sign of increasing lead traction. As the manifestation of a twiddler syndrome evolves over time and clear symptoms and signs may be lacking, prompt follow-up and chest radiography is crucial and should be performed in case of lead-related remote monitoring alerts to avoid complications.

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Fig. 1 Remote monitoring of the RV lead. The mean R-wave amplitude (black) and the pacing threshold (red) are shown

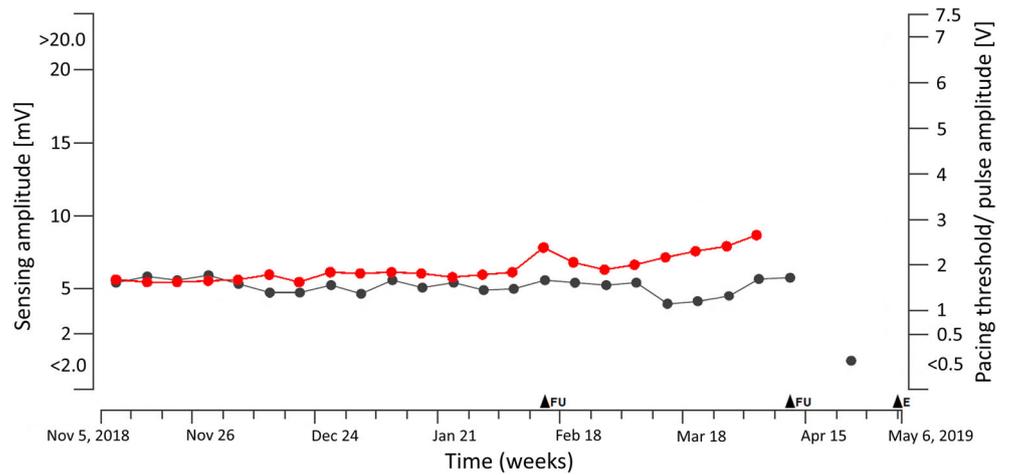
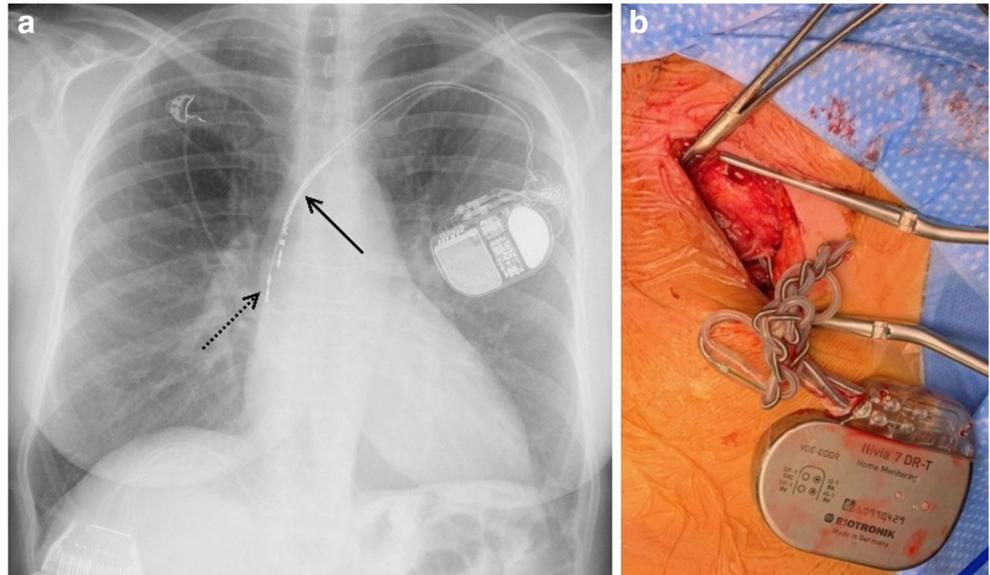


Fig. 2 **a** X-ray showing the dislodgment of both leads (solid arrow, ventricular lead; dotted arrow, atrial lead). **b** Intraoperative situs during ICD revision



Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

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