

CASE REPORT

Unique abdominal twiddler syndrome

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Abstract

Twiddler syndrome is an uncommon complication that occurs by twisting of the generator and may cause torsion, dislodgement, and injury of the leads. We report a rare case of a twiddler syndrome associated with an abdominal permanent pacemaker. Abdominal twiddler syndrome may possess a unique mechanism, which may not be seen in chest twiddler syndrome.

KEYWORDS

abdominal distension, complication, epicardial lead, pacemaker, twiddler syndrome

1 | CASE PRESENTATION

An 82-year-old woman with symptomatic sick sinus syndrome underwent an implantation of a permanent pacemaker. She had a history of a triple valve replacement with the simultaneous implantation of epicardial pacing leads (Intermedics Model: 471-07, 5/6 mm pin) on the ventricle 29 years prior. Those 29-year-old leads were connected to a newly implanted pacemaker (Medtronic Adapta ADDR06) in the abdomen since adequate sensing, pacing, and impedance values were confirmed at the time of the pacemaker implant. Six weeks after the implantation, she presented to our hospital with a complaint of a spontaneous abdominal protrusion and intermittent pain at the pacemaker pocket site. The pacemaker generator spontaneously took a stand-up position in the abdominal pocket regardless of her body posture (Figure 1A: supine posture and Figure 1B: standing posture). Before she presented to our hospital, she repeatedly manipulated the generator from the perpendicular position to the flat position of her abdominal wall in the pocket. Her increasing postprandial abdominal distention made the generator reproducibly and spontaneously restand in a perpendicular position because she had a background of chronic abdominal distention due to functional chronic constipation and intestinal gas.

The X-ray examination revealed multiple small loops of the pacemaker leads, which resembled the twiddler syndrome usually observed on the chest wall (white arrow in Figure 2A). We performed a pacemaker pocket revision to undo the cluster of tangled leads

connected to the device (Figure 2B). The generator was sutured tightly to the fascia at multiple sites of the suture hole and of the pacemaker header grooves with nonabsorbable sutures (Figure 3A). Additionally, the pacemaker pocket was shrunk by suturing the surrounding connective tissue of the pocket (Figure 3B). After the operation, the device rotation and her abdominal pain resolved.

2 | DISCUSSION

Twiddler syndrome is a rare complication caused by spontaneous or self-induced manipulation of the pacemaker pulse generator around its central axis within the pocket, resulting in retraction and dislocation of the leads. It has a reported frequency of below 0.1%, and the majority of cases are diagnosed within the first year of the implant.¹⁻⁴ In most cases, their manifestations depend on the final site of the dislodged lead including the loss of capture, stimulation of noncardiac structures, or inappropriate ICD discharges.¹⁻⁵ In this particular case, a unique abdominal twiddler syndrome occurred because (a) the pacemaker took a perpendicular position spontaneously in the pocket due to chronic abdominal distention and (b) the patient repeatedly manipulated the pacemaker that had stood up in the pocket to a flat position.

Elderly patients have been reported to have an increased risk of chest twiddler syndrome since the presence of loose subcutaneous tissue allows for the rotation of the generator in its pocket.¹⁻⁴

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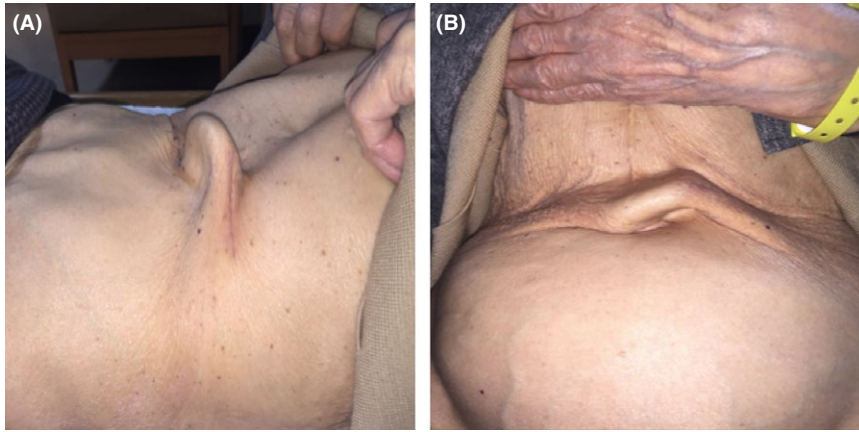


FIGURE 1 Abdominal images after a meal showing the pacemaker generator standing perpendicularly regardless of her posture. A, Supine posture and B, Standing posture. Note that severe postprandial abdominal distension was observed

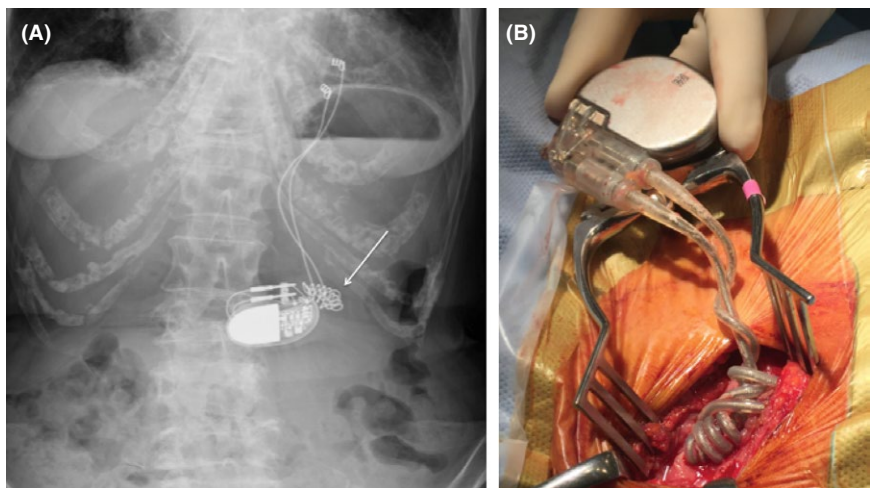


FIGURE 2 A, The chest radiographs showing numerous loops and twists of the pacemaker leads (White arrow). B, Intraoperative image showing the cluster of tangled leads connected to the device

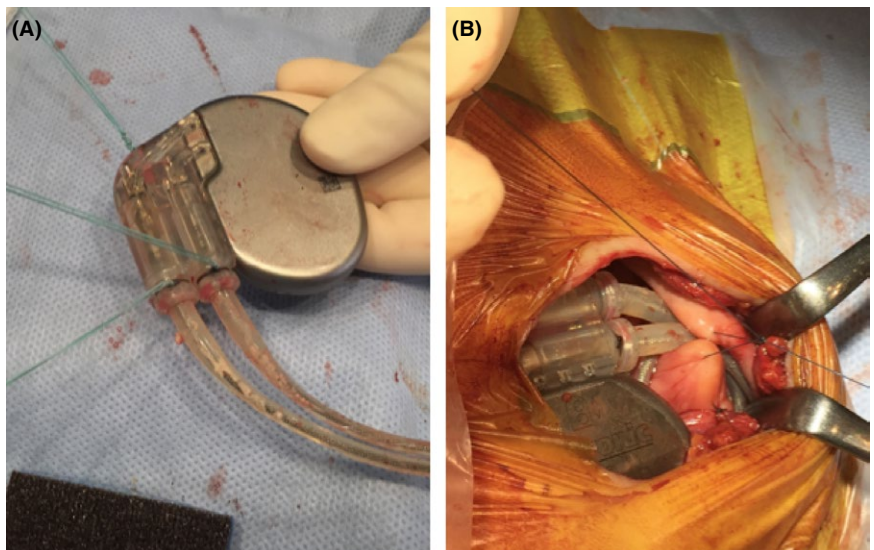


FIGURE 3 A, Intraoperative image showing the multiple suture sites for fixing the pacemaker tightly in the pocket. Note that not only the suture hole but also the grooves in the two header connector ports are sutured with nonabsorbable sutures. B, Intraoperative image showing that the size of the pacemaker pocket had shrunk. Note that the connective tissue in the pocket was sutured

Furthermore, a smaller sized implanted device relative to its pocket may also present an increased risk by providing the space needed for a greater displacement of the device within the skin pocket.^{4,5} The risk of abdominal twiddler syndrome might be similar. However, the biggest difference between abdominal implantations and chest implantations is the stability of the pacemaker pocket base. Although the chest wall, which consists of bones and muscles, is stable and the cause of chest twiddler syndrome is patient error, the abdominal twiddler syndrome we reported was caused by the patient's physical elements.

3 | CONCLUSION

In the case of abdominal twiddler syndrome, postprandial abdominal distension, which is not a risk of chest twiddler syndrome, may be a possible risk for its incidence.

CONFLICTS OF INTERESTS

Authors declare no conflict of interests for this article.

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How to cite this article: Higuchi S, Shoda M, Satomi N, et al. Unique abdominal twiddler syndrome. *J Arrhythmia*. 2019;35:142-144. <https://doi.org/10.1002/joa3.12133>