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Absorbable antibacterial envelope in the surgical management of Twiddler's syndrome in a patient with gastric electric stimulator: a case report

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Abstract

Gastroparesis is a chronic gastric motility disorder characterized by delayed gastric emptying and a multitude of troublesome symptoms, including chronic nausea, vomiting, abdominal pain, malnutrition, and dehydration. Whereas initial management of the gastroparesis is conservative, patient with refractory gastroparesis may benefit from surgical therapy, including gastric electric stimulator (GES) device implantation. Twiddler's syndrome is a challenging condition well described in the cardiac literature that is characterized by the instability, displacement, leads twisting and resulting malfunction of an implanted device, believed to be due to manipulation (twiddling) by the patient. The condition is not specifically characterized in the GES literature; however, evidence suggest the incidence of the Twiddler's syndrome is reaching up to 9% of the patients with GES. In the current report we present a case of surgical management of the recurrent Twiddler's syndrome in a patient with a GES device with novel non-FDA approved use of the TYRX™ Absorbable Antibacterial Envelope for the device stabilization. Use of the TYRX™ enveloped provided additional anchoring points of fixation, resulting in the successful resolution of the device instability and the Twiddler's syndrome with and ongoing follow up of up to 8 months. This report adds to the armamentarium of surgical management of this uncommon and difficult problem.

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Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this case report were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent for the publication was obtained from the patient.

Keywords

Absorbable antibacterial envelop; gastric electric stimulator (GES); Twiddler's syndrome; surgical revision of the implantable device

Introduction

Gastroparesis is a chronic gastric motility disorder characterized by delayed gastric emptying and a multitude of troublesome symptoms, including chronic nausea, vomiting, abdominal pain, malnutrition, and dehydration. Initial therapy for gastroparesis is conservative with dietary changes and pharmacologic therapy, particularly anti-emetic and prokinetic agents. Surgical interventions, such as pyloric drainage procedures or gastric electric stimulation typically reserved for patients with refractory gastroparesis (1–6).

Twiddler's syndrome, the excessive device mobility and resulting malposition and malfunction of a cardiac pacemaker was first described in by Bayliss *et al.* in 1968 (7). The excessive device mobility in Twiddler's syndrome is believed to result from manipulation of the pulse generator by the patient, causing rotation of the device with coiling, dislodgement and even fracture of the leads. The syndrome is relatively uncommon, with an estimated frequency between 0.07–7% (8,9). A well-recognized scenario in the cardiac literature, it has not been specifically characterized in patients with gastric electric stimulators (GES). We describe a case of successful surgical management of the recurrent Twiddler's syndrome in a patient with GES with the use of antibiotic impregnated mesh envelope for the additional fixation and stabilization of the device. We present the following article in accordance with the CARE reporting checklist (available at http://dx.doi.org/10.21037/dmr-20-70).

Case presentation

A 53-year-old obese woman with refractory diabetic gastroparesis underwent re-implantation of a GES 14 months after explantation of a previous device due to need of MRI. Initially she was doing well until her symptoms of nausea, vomiting and abdominal pain have returned 4 months postoperatively. Interrogation of the device revealed abnormal impedance values. Abdominal X-ray confirmed malpositioned device with twisted and broken wires (Figure 1), consistent with Twiddler's syndrome.

Revisional surgery revealed twisted and broken leads, that were replaced (Figure 2A). The device was secured with an additional transfascial suture over the middle of the device body, assuring satisfactory device immobilization (Figure 2B). At the initial post-operative visit the device was functioning appropriately with significant symptom improvement.

Four months after revisional surgery the patient reported again recurrence of gastroparesis symptoms, shocking sensation and device mobility. Abdominal radiograph confirmed displacement of the device and twisting of wires, consistent with recurrent Twiddler's syndrome (Figure 3). On physical examination the device could be easily manipulated and flipped in the pocket.

The patient underwent repeat surgical revision. TYRX antibiotic impregnated mesh envelop (Medtronic, Minneapolis, NM, USA), was selected for additional device anchoring (Figure 4A). Open surgical exploration revealed entwined but intact leads with normal impedance values (Figure 4B). The device was found to be loose and freely rotating in a large patulous pocket without any adhesions. The leads were untwined and assured intact. After electrosurgical ablation of the pocket lining, the device was fitted with the envelope and secured with the two standard transfascial Prolene sutures. Two additional transfascial perpendicular horizontal mattress-type running Prolene sutures, incorporating the TYRX envelope, were used to firmly secure the device to the underlying fascia (Figure 4C). The patient tolerated the procedure well with resolution of her symptoms. At three months postoperatively there was no evidence of device mobility with stable position of the device on imaging (Figure 5). Patient remains asymptomatic at the time of publication with 9 months follow up. All procedures performed in this case report were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent for the publication was obtained from the patient.

Discussion

GES devices were approved by the FDA under the Humanitarian Use Device (HUD) exemption in 2000 as a treatment for refractory idiopathic or diabetic gastroparesis (6,10). The GES device consists of a pair of electrodes that are inserted into the gastric muscularis propria along the greater curvature of the stomach and connected to the pulse generator, implanted in a subcutaneous pocket of the abdominal wall.

Although not specifically characterized, there is evidence in the literature to suggest that Twiddler’s syndrome is present in patients with GES devices. Early studies of GES devices found that common adverse effects included device migration and flipping, that occasionally occur due to patients manipulating the device (1,11,12). Superficial pocket location, particularly in patients with obesity, was associated with increased rates of device malposition and increased rates of surgical revision of GES devices (1,10,11). In the report on surgical outcomes of 233 patients after GES implantation, 21 (9%) required surgical revision of the subcutaneous pocket. Logistic regression revealed 9% increase in risk of revision for each unit of BMI increase. Obese patients had an overall 4.5 times increased risk of posket revision (11). A lack of the abdominal wall stability might be a contributing factor for the development of the abdominal Twiddler’s syndrome (13). Additional adverse effects, such as electric shock sensations, might further suggest an unidentified Twiddler’s syndrome causing the damage of plastic lead covering and might require surgical revision or device explantation (1,10).

Surgical revision is a mainstay of the treatment of the Twiddler’s syndrome. Some patients may require multiple device revisions as they often continue to twiddle the devices postoperatively (8). Most patients deny manipulation of the device, though cases of deliberate manipulation attributed to inadequate patient education have also been reported (8). Some authors believe that an inadequate fixation of the device, leading to relatively free rotation within a loose and capacious subcutaneous pocket, is the primary cause of

Twiddler's syndrome. Patient attempts to return the rotated device into the normal position is a secondary contribution to the problem (14).

Use of a synthetic pouch for prevention of Twiddler's syndrome in a patient with automatic implantable cardioverter defibrillator (AICD) was first described in 1995 (14). In a retrospective analysis of 21 patients with Twiddler's syndrome use of nonabsorbable polypropylene envelopes completely eliminated recurrent events compare to 50% incidence of re-twiddling prior to the use of the envelopes (15). Minimizing pocket size and suture fixation of the pulse generating device have also been described to limit the occurrence of the Twiddler's syndrome (6). More recently, the use of absorbable antibiotic mesh envelopes has been proposed in cases of Twiddler's syndrome in patients with cardiovascular implantable devices (16). The TYRX™ is an absorbable antibacterial mesh envelope with minocycline and rifampin, that is FDA approved for use in patients with higher risk of cardiovascular implantable electronic device (CIED) infection. In our case, use of the TYRX™ envelope provided additional anchoring points, resulting in successful surgical revision of the recurrent Twiddler's syndrome in the patient with GES. To our knowledge the use of the TYRX envelope has not been described in patients with GES Twiddler's syndrome.

This current publication is a first case report, based on the single experience of successful surgical treatment of the recurrent Twiddler's syndrome and as such presents a limited evidence for the management of the described problem. However, success in resolution of this rare and arduous problem adds to the armamentarium for the surgical management of this challenging condition.

Conclusions

We describe a non-FDA approved use of the TYRX™ Absorbable Antibacterial Envelope for the management of recurrent Twiddler's syndrome in a patient with a GES device. The TYRX™ envelope provides extra fixation points for the anchoring of the prolene suture on the GES device, allowing superior device stability and successful surgical treatment of recurrent Twiddler's syndrome.

Acknowledgments

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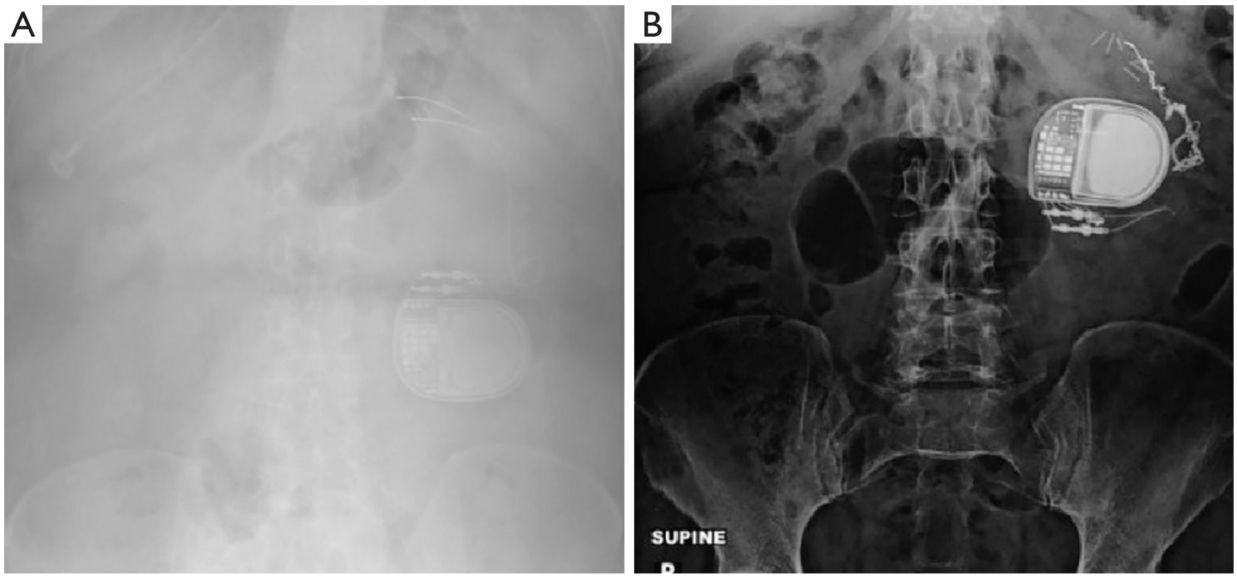

Figure 1.

Abdominal imaging documenting device and leads position. (A) Immediately postoperative (Jan 2019). (B) 4 Months postoperative (May 2019). Please note flipping of the device and twisting and fracture of the leads.

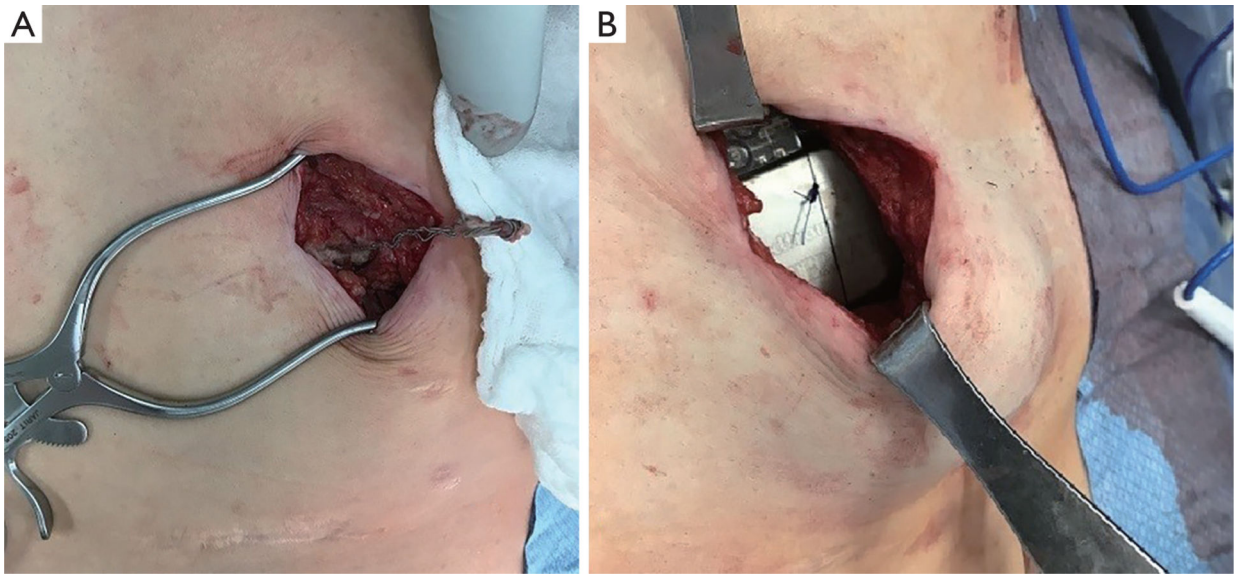

Figure 2.
Intraoperative findings during first revisional surgery (June 2019). (A) Twisted and fractured leads. (B) Fixation of the device with an additional fascial stitch over the middle of the device body.

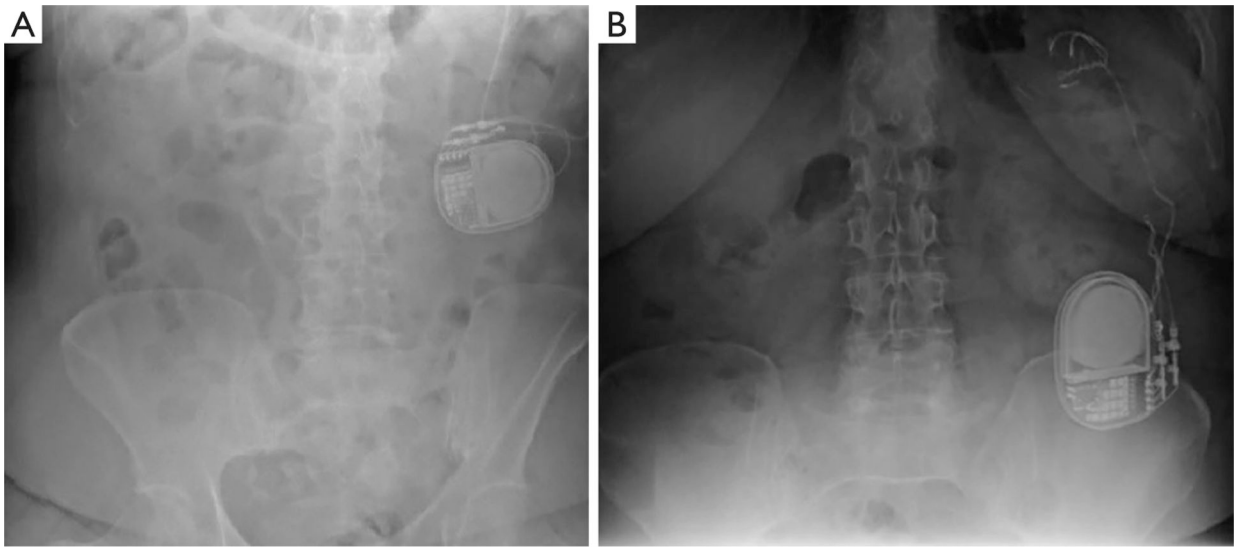

Figure 3.
Abdominal imaging documenting position of the device and leads. (A) Imaging after first revisional surgery (June 2019). (B) Abdominal imaging 7 months after first revisional surgery (Please note flipping of the device with twisting of the leads).

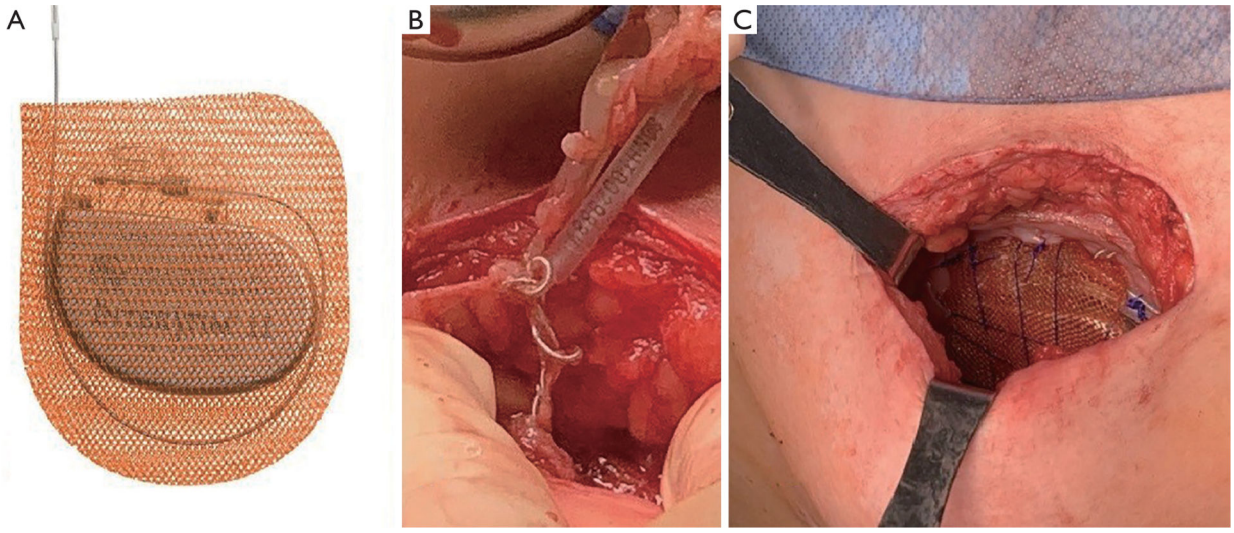

Figure 4.

Intraoperative findings during second revisional surgery with implantation of the TYRX antibacterial envelope. (A) TYRX antibacterial envelope. (B) Intraoperative appearance of the wires during second revisional surgery. (C) Securing of the device with Prolene stitch, incorporation TYRX envelope.

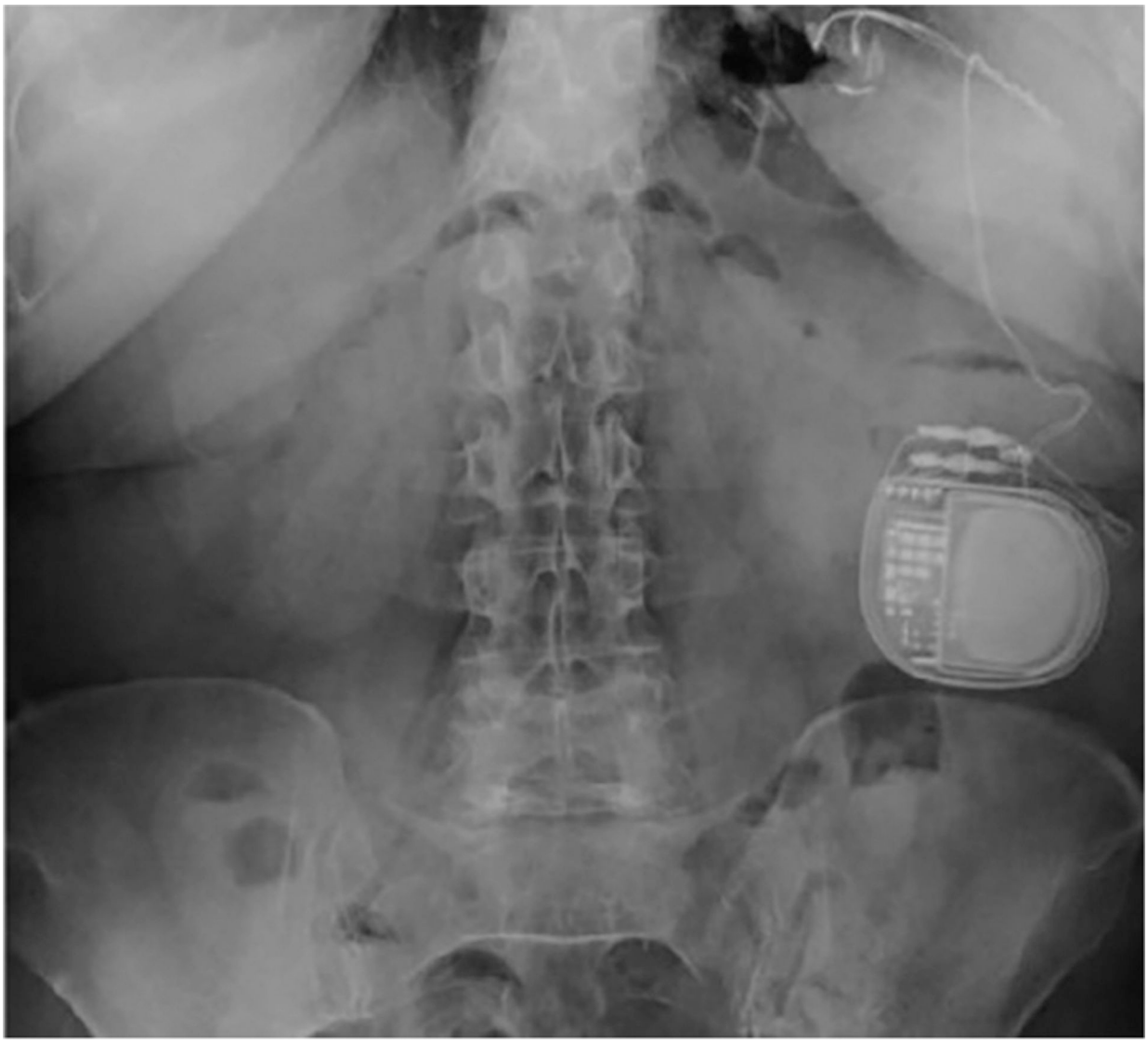

Figure 5.
Stable imaging after second revisional surgery (March 2020)—persistent twining of the non-replaced intraabdominal portion with normal course of the subcutaneous portion of the leads. Device remains in stable position and functioning appropriately.