

Twiddler's syndrome in a sacral nerve stimulator

A 68-year-old female presented with return of passive faecal incontinence 6 months post-insertion of a sacral nerve stimulator (SNS). She previously had a laparoscopic ventral rectopexy in 2020, haemorrhoidectomy and transanal Delorme procedure for mucosal recurrence of her rectal prolapse 2021. She had a BMI of 32, was a non-smoker, and an active farmer. She was on no regular medications. After SNS implantation, there was significant improvement of faecal incontinence and urgency but this had stopped working at 3 months. Despite trialling all the programs at maximum amplitudes, she had no buttock or lower limb paresthesia. An x-ray showed migration and spiralling of the SNS wires outside the sacral foramina (Fig. 1). Subsequent removal and replacement of the SNS showed twisting of the wires along its longitudinal axis (Figs 2 and 3). The patient denied any recent trauma or 'twiddling' the device. The new SNS worked well.

Twiddler's syndrome is a well-known phenomenon in patients with cardiac pacemakers and implantable cardioverter-defibrillators.¹ Less commonly, it has been reported in those with chemotherapy infusion pumps, deep brain stimulators, vagal nerve stimulators and spinal cord stimulators.^{1,2} However, Twiddler's syndrome has never been reported in SNS.

Pacemaker associated Twiddler's syndrome was first reported in 1968 due to patient manipulation of the pacemaker device causing coiling of lead wires along its long axis.³ Typically twisting of

wires causes lead retraction and displacement, lead fracture and risk of life-threatening arrhythmias.⁴ Rotation of wires along the short axis is known as "Reel syndrome."²

Twiddler's syndrome occurs in 0.39–1.3% of device implants but is likely under-reported and under-recognized.^{1,2} The exact pathogenesis is unclear but is attributed to patient conscious or subconscious twiddling of the device, contraction of the underlying muscle and a proportionally larger pocket to device ratio.² Risk factors include obesity, elderly, female, psychiatric disease, within the first year of device placement and weight loss.^{2,4–6}

In SNS, a subcutaneous pocket is made just deep to Scarpa's fascia in the upper lateral buttock and away from the ischial tuberosities. Leads are tunnelled from the third sacral foramina to the subcutaneous pocket via the paramedian sacral needle insertion site. Unlike cardiac pacemakers which have a subpectoral pocket and a bony rib cage for support, the pocket for the SNS battery is entirely subcutaneous and superficial to the gluteus muscles, which may predispose to rotation.² In addition, patients with SNS can adjust the stimulation intensity and programming by placing a device over the battery which may lead to increased patient awareness and manipulation of the battery.

Reported techniques to decrease the risk of Twiddler's syndrome include suture fixation of the device, use of a pouch or

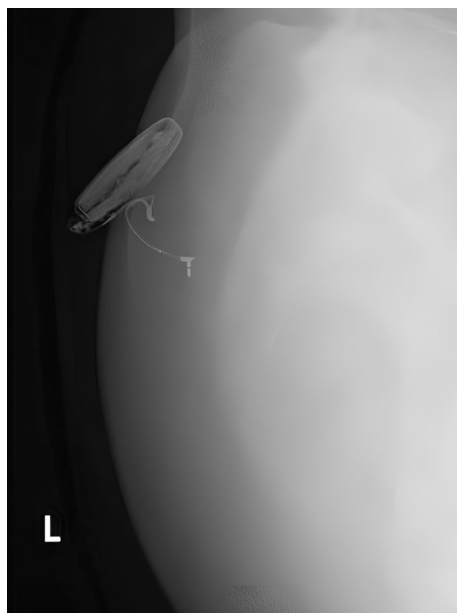


Fig. 1. Lateral x-ray of sacral nerve stimulator with retracted and coiled leads.

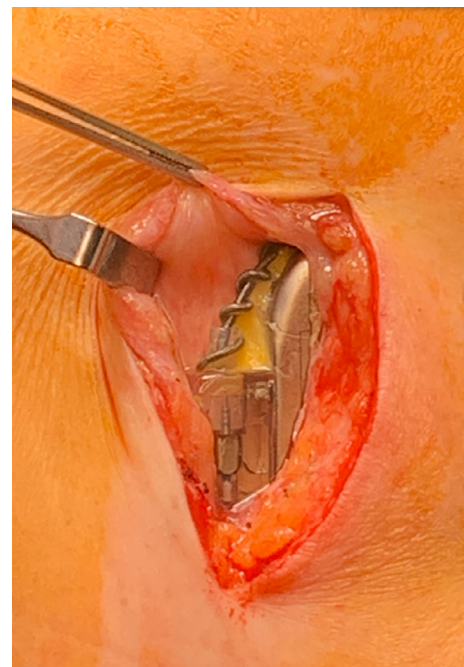


Fig. 2. Sacral nerve stimulator in subcutaneous pocket with coiled wires.

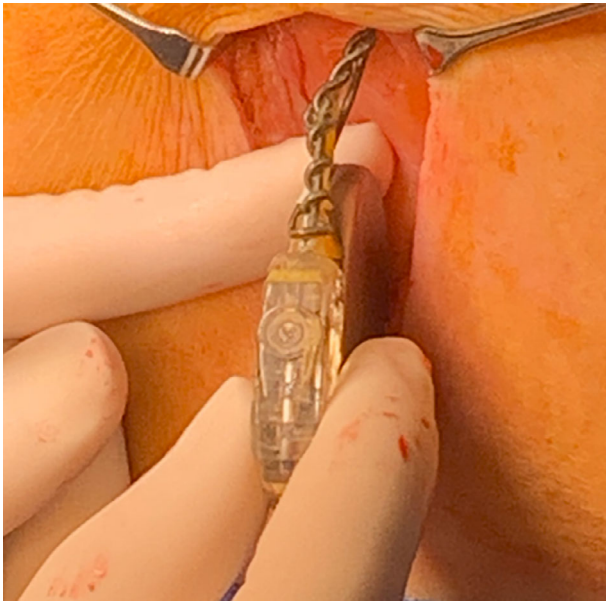


Fig. 3. Sacral nerve stimulator battery with coiling of wires along its longitudinal axis.

mesh to promote tissue fixation, compression dressings to decrease risk of seroma permitting dilation of the pocket and creation of a tight pocket.²

Twiddler's syndrome is a rare but likely under-appreciated cause of SNS malfunction and should be considered during troubleshooting of malfunctioning SNS.

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Author contributions

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