TWIDDLER’S SYNDROME AFTER DORSAL ROOT GANGLION STIMULATOR IMPLANTATION: A CASE REPORT AND REVIEW OF THE LITERATURE

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Introduction
Twiddler’s syndrome is a rare hardware complication of implantable pulse generators (IPG), associated with repeated coiling and twisting of the IPG and leads¹. The first description of the syndrome dates 1968 when Bayliss et al. described it in cardiac transvenous pacemakers. Since this first description, the syndrome has been well documented in cardiac pacemakers and deep brain stimulators²⁴. In the last years, twiddler’s syndrome has also been described in patients with spinal cord stimulators³⁵ and with the increase in cases, more and more interest has been invested in understanding the pathophysiology of the syndrome. To our knowledge, we report the first case of a patient with dorsal root ganglion stimulation developing Twiddler’s syndrome and review the current literature regarding this pathology.

Case presentation
A 51-year-old woman with complex regional pain syndrome of the left foot and calf after malleolar fracture received dorsal root ganglion stimulation with implantation of two electrodes on the left-sided L5 and S1 nerve roots. The IPG was implanted abdominally on the right side. Postoperatively the patient showed reduction of pain from NRS 8-9 to 5. Over the following months, she developed increasing discomfort along the connector and extension leading to the IPG. 4 months postoperatively, the patient reported severe pocket pain, as well as pain along the electrode passage. Fluoroscopy showed strain of the electrodes with entanglement of the leads (Figure 1). Revision surgery was planned, and the patient wished to have the IPG implanted lumbar on the right side. Intraoperatively, the extension leads were completely entwined and twisted, resulting in extensive strain on the connector and IPG (Figure 2). Disconnection of the extensions leads from the battery showed, that the DRG-electrodes had been damaged by the strain with the distal contacts of the leads torn out (Figure 3). The electrodes were removed and replaced. After surgery the patient described complete resolution of her pain.

Discussion
Twiddlers syndrome has been described in up to 7% of patients with cardiac pacemakers² and 1.5% with DBS³. The first description of this pathology in patients receiving a spinal cord stimulation was made by Al-Mahfoudh et al. in 2016⁴ with an incidence of 0.5%. Despite growing reports in the literature, the pathophysiology behind this syndrome remains unclear. Deliberate or subconscious manipulation of the device as well as the result of muscle contraction on the IPG are possible mechanisms. Old age, female gender, obesity, abdominal site of implantation and psychological disorders have been described as possible risk factors in the development of this syndrome. Our patient was female, obese with a BMI of 32kg/m² and had initially her IPG implanted abdominally, all of these being described as risk factors for developing Twiddlers syndrome⁵.

Conclusions
To our knowledge, we report the first case of Twiddlers syndrome with implantable pulse generators for dorsal root ganglion stimulation. As in spinal cord stimulation, cardiac pacemakers and deep brain stimulation, the awareness of this syndrome may prevent its occurrence. Especially in obese patients, implantation of an IPG abdominally puts the patient at risk for developing Twiddler’s syndrome, so that IPG-placement in the lumbar region should be considered as the first implantation site.

References