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## Obsessive Compulsive Disorder in a Patient with Twiddler's Syndrome

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**Abstract**

*Twiddler's or Twist syndrome is the twisting of pulse generators around themselves. It may result from mechanical manipulation that can induce the malfunction of the device. In this case, twiddler's syndrome resulted from compulsive checking of the device. The implantable cardioverter-defibrillator (ICD) triggered the development of an obsessive compulsive disorder (OCD). Two invasive procedures were required to replace the ICD. Psychiatric intervention prevented the recurrence of twiddler's syndrome in this patient for more than two years. We believe that pre-implant psychiatric assessment should be the rule.*

***Twiddler's syndrome, obsessive compulsive disorder, anxiety, cognitive behavior therapy***

## **Background**

First described by Bayliss et al. in 1968 [1], twiddler's or twist Syndrome is the twisting of pulse generators around themselves. A total or partial movement of the pulse generators or even a rupture of the isolating material around the generators may be responsible for the malfunction of the device. The etiology of the syndrome is often considered to be a large subcutaneous pocket of abundant fatty tissue in obese subjects. It may affect a pacemaker or an implantable cardioverter-defibrillator (ICD) and can be easily diagnosed on a chest X-ray. Re-intervention consists in the replacement of the pulse generators and the creation of a tight generator pocket.

The present report focuses on its psychological causes in a case due to post-implant compulsive behavior, resulting in two interventions to replace the ICD. The essential features of obsessive-compulsive disorder (OCD) are recurrent obsessions and compulsions severe enough to be time-consuming and to cause marked impairment.

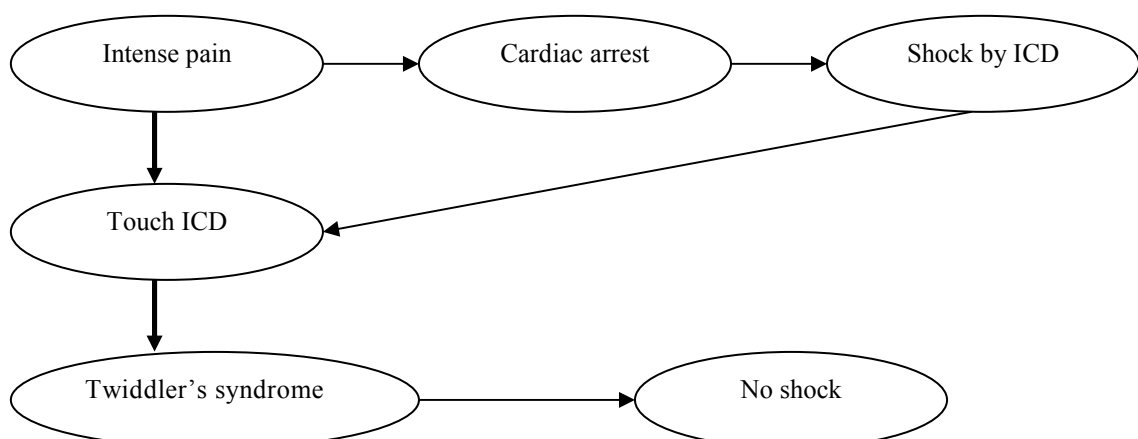
## **Case History**

A 47 year old male smoker with hyperlipidemia, prior history of myocardial infarction (MI) and subsequent ventricular fibrillation underwent implantation of an internal cardioverter defibrillator (ICD). The location of the ICD was in the left infraclavicular area beneath the pectoral muscle. The patient was referred for psychiatric consultation after requiring 3 revisions of the ICD system for lead displacement.

On assessment, he had no previous psychiatric history but was noted to have obsessional personality traits. At home he had been preoccupied with order and cleanliness resulting in regular arguments with his wife. Following the chest pain associated with his first MI 10 years ago, he believed that any subsequent chest pain heralded a further MI with the risk of death. Since the implantation of the ICD 16 months previously, the patient had

increased anxiety and described fears of having chest pain and of dying as well as of receiving ICD shocks. He had developed compulsive checking behavior focused on the ICD and would touch his chest repeatedly up to 50 times/day. He stated that touching his chest helped to ease his pain and also allowed him to check that the ICD was in place. Over time, he developed a precise way of touching the defibrillator which involved hooking the 4 fingers of his right hand in his left armpit whilst moving his thumb on the defibrillator from right to left. These compulsive actions explained how the ICD had turned on itself and the pulse generators had become twisted. In brief, this patient appeared to have developed an anxious cognitive construct that resulted in him checking the device compulsively to reduce his fear of death or receiving an ICD shock (see diagram below: briefly, pain was interpreted as a cardiac arrest, necessitating an ICD shock. This need, together with the associated need to alleviate the pain, resulted in the repeated touching of the device, in turn triggering the twiddler's syndrome). He scored 16/40 on the Yale-Brown Obsessive Compulsive Scale (Y-BOCS) [4] reflecting a moderately severe OCD [5].

**The patient's cognitive construct was as follows:**



The patient received Cognitive Behavioral Therapy (CBT) involving exposure with response prevention (ERP) combined with an antidepressant (escitalopram 20 mg per day) [6]. Exposure method consisted of prolonged exposure in imagination to the feared stimulus (anxious thoughts or image) and interoceptive exposure to chest pain until anxiety subsided. At the same time, the patient was instructed not to engage in any checking behavior. A qualified psychiatrist (NJ) administered individual CBT. Sessions took place for one hour once a week over 5 months, then twice a month during 6 months, with a total of 32 sessions over 11 months. In addition, 15 sessions of Schultz relaxation exercises were given which use visualization techniques to induce relaxation.

No signs of recurrence of the syndrome were noted for 3 years. At the end of the therapeutic period, the Y-BOCS score dropped to 3/40 (No OCD).

- Please insert here Figure 1 -

### **Discussion**

This case clearly illustrates the role of compulsive device-checking behavior in the etiology of twiddler's syndrome. In this patient with an obsessive personality, the ICD became the trigger for his current OCD. Three defibrillators had to be replaced. CBT with relaxation sessions and antidepressant treatment led to improvement of the OCD and prevented relapse of twiddler's syndrome.

Twiddler's syndrome is considered a rare complication. In 1989, Solti et al. [7] reported 6 cases in 4250 implants. In 1994, a literature review revealed only 10 reported cases [8]. However, Viard [9] suggests that underestimation of prevalence may occur on account of a lack of movement of the pulse generators or because of the rupture of the

isolating material around the generators, or because chest X-rays are not performed systematically. The syndrome usually occurs between 16 days [10, 11] and 36 months [12] after insertion of the device. In this case it occurred 16 months after implantation. Typically, it is commoner in women and affects obese subjects with little muscular tissue and abundant subcutaneous fatty tissue, and with some psychiatric background (e.g., generalized anxiety disorder). To date, two types of etiological factors have been established: mechanical [13-16] (large subcutaneous pocket in obese women with abundant fatty tissue, [7, 10]) and psychiatric (manic syndrome) [7, 8, 17]. The insertion technique itself has not been implicated. This is the first report of OCD as an etiological factor.

Beauregard et al. [18] reported the case of a 44-year-old patient with an endocavitary pulse generator and a subcutaneous patch who, after losing 30kg in weight, developed twiddler's syndrome, resulting in six shocks. It was not clear however whether the weight loss was related to a pre-existing psychiatric condition or was secondary to the cardiac illness. Furman [19] suggested the syndrome was more frequent when the stimulator pocket is larger than the apparatus. To counteract this, a Parsonnet dacron pocket may be used. However, Avitall et al. [20] reported the occurrence of the syndrome despite using the pocket. Since fixing the stimulator to the bottom of a smaller pocket does not always prevent the occurrence of twiddler's syndrome [17], evaluating for a psychiatric cause may be appropriate.

Very often patients report not touching their device. This notion of the « unconscious or sub-conscious manipulation » has already been reported [8, 17]. The phenomenon could be related to the patient's « degree of insight », which is the level of recognition of their illness. Sometimes patients touch the apparatus in the early period following implantation, as an automatic reflex to alleviate the pain due to the scar and to check the integrity of the device as an adaptive behavior to the foreign object. Quite often,

patients give it a nickname (e.g., my baby, my toto, my rescuer, etc.) in order to accept it psychologically. While such manipulations may cease quickly in patients with no psychiatric history and a good level of insight into their cardiac illness, they may continue in those suffering from a psychiatric disorder and/or with poor insight into their illness. In the present case, the patient had good insight but showed marked obsessional personality traits.

The latest defibrillators are smaller, more efficient and multifunctional. Reporting a patient who developed twiddler's syndrome following insertion of a small ICD, Crossley et al. [21] suggested that the small size of the ICD, its retro-pectoral position and the resultant discomfort may be responsible for triggering the syndrome. However, no data were provided on the personality or psychiatric history of the patient.

Twiddler's diagnosis is confirmed by a chest X-ray. Reinsertion is generally necessary with the replacement of the pulse generators and the creation of a smaller pocket. A recent study recommended regular chest x-ray follow-up to enable early diagnosis and treatment of twiddler's syndrome [22]. Others suggest that, in addition, routine magnetic tests should be administered [9]. The present case also shows clearly that pre-implant psychiatric assessment is important as this can help identify psychological/psychiatric factors which may contribute to the etiology of twiddler's syndrome. Anxiety and stress may lead to compulsive behavior with its dysfunctional consequences. Hence, CBT and stress management may reduce anxiety, have positive effects on stress-induced fibrillation [23, 24] and prevent twiddler's syndrome. This needs to be confirmed by systematic studies. In clinical practice, a biopsychosocial approach, which includes a psychiatric assessment [25], can identify patients at risk for twiddler's syndrome. Our case study suggests that psychological interventions may decrease this risk.



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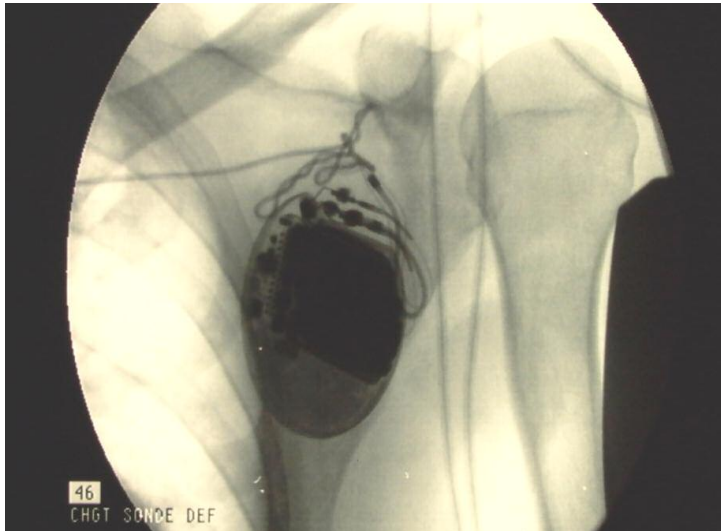


Photo of Twiddler's syndrome