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Case report

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Reel's syndrome, an unusual cause of lead displacement

El síndrome de Reel, una causa inusual de desplazamiento de electrodos

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ABSTRACT

Background: Ventricular capture failure could be a sign of macrodisplacement. Electrode displacement is one of the complications related to cardiac pacing. Knowing the different production mechanisms of macrodisplacements is not just for a nominal distinction. It is important to understand that the mechanisms are different, that´s why each type of macrodisplacement requires different form of therapy.

Case report: A 79-year-old male patient. On physical examination, the heart rate was 38 beats per minute. The 12-lead electrocardiogram (ECG-12) showed a second-degree 2:1 atrioventricular block. It was decided to implant a VVIR pacemaker. On follow-up, the ECG-12 review showed ventricular capture failure without spike and 2:1 atrioventricular block. The fluoroscopic study showed electrode displacement. The distal tip of the electrode was in the area of the pacemaker pocket with radiological appearance of thread wound on a reel. Reintervention was immediately decided, during surgery, the diagnosis of reel type macrodisplacement was corroborated. The lead was repositioned in the right ventricle.

Conclusions: Knowledge of the risk factors allows us to avoid macrodisplacement syndromes of the electrode. Knowing the different mechanisms of production of the three syndromes described is of great importance to finally carry out the appropriate therapy**.**

Keywords: Macrodisplacements; reel syndrome; Ratche syndrome; Twiddler; Ventricular capture failure

1. Background

Electrode displacement is one of the complications related to cardiac pacing. Typically, it occurs in two periods after implantation of cardiac pacing systems; early most often within the first 6 weeks or late beyond 6 weeks (Chauhan *et al.,*1994). Classically the macrodisplacements or macrodislocations, have been described and called twiddler syndrome. Other authors, also include the iatrogenic displacements or displacements produced by trauma on the pacing system (Bayliss *et al,*1968; Carnero-Varo *et al.,* 1994).

There is a "knowledge gap" based on the fact that although the concepts of the different macrodisplacements are currently well defined, there are several cases reported in the literature under incorrect nomenclature. Statistics may include biases because identical cases reports have been defined and classified differently at the authors' criteria and, conversely, different cases have been classified in the same way. Thus, cases initially described as twiddler syndrome are actually forms of reel syndrome, and cases described as either of these two entities would actually represent ratchet syndrome (Bracke et al,2005; Higgins et al., 1998).

In this sense, it is common to find in the literature cases reports of macro-displacements whose mechanism differs of the twiddler's syndrome. However, they are described under this denomination (twiddler's syndrome) when in fact the forms of presentation are different. Those mentioned above with different form of presentation should be included within the macrodisplacement syndromes with another name (Mahdi *et al.,*2023).

Knowing the different production mechanisms of macrodisplacements is not just for a nominal distinction. It is important to understand that the mechanisms are different, that´s why each type of macrodisplacement requires different form of therapy. Those previously recognized as variants of Twiddler syndrome are currently defined with a well-established mechanism of production of electrode macrodisplacement.

An updated synthesis or summary to understand the different types of macrodisplacements and their implications was not found in the literature reviewed; and even, in the opinion of the authors, there are inconsistencies in some published reports, the objective of the present work was to describe a clinical case accompanied by a documentary review of the different syndromes of macrodisplacements.

2. Case presentation

Once the patient has signed the informed consent form, in order to make the case report, we requested the approval of the ethics committee of our center, taking into account the principles of beneficence and non-maleficence in order to transmit in the literature the knowledge that this report can spread in medical sciences.

A 79-year-old male patient, obese, with a previous history of hypertension and type 2 diabetes mellitus. He came to the cardiology department for consultation, reporting marked decline on minimal

exertion. On physical examination, heart rate was 38 beats per minute. The 12-lead electrocardiogram (ECG-12) showed second-degree atrioventricular block type 2:1. VVIR pacemaker implantation was decided and the patient was discharged after the procedure (in this case, BIOTRNIK's INTICA generator was used for permanent pacing, without programming defibrillation functions.

It was a recovered and resterilized generator because we are from a low to middle-income country and we did not have a stimulation generator at this moment and it was necessary to save the patient's life). In the follow-up, established for the first and third month after pacemaker implantation, it was possible to verify the existence of electrical parameters within the normal range and the thoracic Rx showed in those two moments the correct location of the device and the electrode. Also, the examination of the pacemaker pocket showed no signs of alterations.

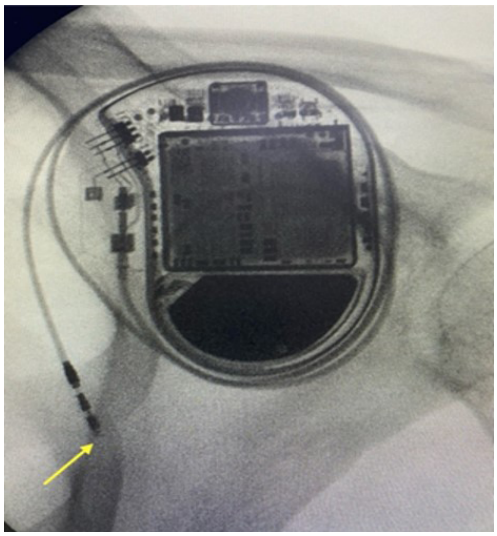
Follow-up at six months: ECG-12 showed ventricular capture failure without spike and 2:1 atrioventricular block. Upon attempting to reprogram the pacemaker parameters, ventricular capture was not achieved despite output voltages of 7.5 volts and pulse widths of 0.6 milliseconds.

The patient was transferred to the electrophysiology and cardiac stimulation lab. The fluoroscopy study showed of electrode displacement. The distal tip of the electrode was in the pacemaker pocket area as shown in figure 1. The reintervention was immediately decided and upon opening the pacemaker pocket the electrode was observed completely rolled up in the direction of the transverse axis in relation to the generator, similar to a thread on a spool; the active fixation attachment of the catheter tip was completely deployed and outside vascular structures. The electrode was reimplanted in the right ventricle by Seldinger technique through the left subclavian vein. The electrode was fixed with Polyester 2.0 suture and the fixation point of the generator was applied to the underlying fascia with the same suture thread. The patient was discharged with normal electrical parameters (ventricular electrode impedance: 820Ω, threshold: 0.9 V).

An important aspect of the present case report is that reel-type macrodisplacement occurs more frequently in the first month after implantation. Our patient presented macrodisplacement at six months. It should be mentioned that the weight of the generator may have had an influence in addition to other factors such as morbid obesity

Fig. 1.

Intica ICD generator (implanted as a pacemaker), Biotronik solia 60 ventricular pacing lead around the generator with an appearance reminiscent of a spool of thread on a reel. The tip of the catheter is seen completely outside the radiological position of the left subclavian vein (below the clavicle) and the fixation mechanism is fully deployed (yellow arrow).



3. Discussion

Twiddler's syndrome is the prototype most often described in the literature; it has an incidence ranging from 0.07- 7% according to some series (Gomez et al., 2023). However, statistics may include biases because identical cases reports have been defined and classified differently at the authors' criteria and, conversely, different cases have been classified in the same way. Thus, cases initially described as twiddler syndrome are actually forms of reel syndrome, and cases described as either of these two entities would actually represent ratchet syndrome (Bracke *et al,*2005; Higgins *et al.,* 1998).

The main gap identified in the literature is the degree of confusion in which identical cases have been reported with different names of macrodisplacements. On the other hand, cases of Reel or Ratchet have been reported as twiddler. There is confusing terminology that may convey errors in teaching in several of the above case reports. For example, Mahdi et al, published a case of twiddler and radiologically it is reel, the same occurred in the report of Nicholson in 2003 and Navarro Gonzalez in 2024. This shows that, although there is a good terminological definition, at present, confusion of terms continues (Mahdi et al, 2023; Nicholson, 2003; Navarro González et al., 2024).

According to the above, it is necessary and essential to define each of these types of presentation of macrodisplacement since their differentiation may have clinical and therapeutic implications. (Arias *et al.,*2012) propose a terminological definition that we believe is appropriate to offer below:

- Twiddler syndrome: retraction and dislocation of electrodes, due to the rotation of the device generator around the axis defined by the electrode (longitudinal axis). Although external manipulation by the patient may facilitate its occurrence, this would not be a necessary condition. Due to the rotating movement of the generator, the electrode would be wound like a braid, allowing a characteristic and defining appearance to be observed.

- Reel syndrome: retraction and dislocation of electrodes due to the rotation of the generator about its sagittal (transverse) axis, which causes the electrode to coil like a reel above or below the generator. Due to the mechanism at work in both Twiddler and Reel syndrome, all electrodes should be affected to a greater or lesser extent, if there is more than one.

- Ratchet syndrome: retraction and dislocation of electrodes caused by the progressive displacement of the electrodes by their fixing parts or their protections, facilitated by the movements of the ipsilateral arm and due to incomplete fixation of the electrode. There is no rotation of the generator about any of its axes. In this case, all the electrodes of a system could be affected or, what may be more common, only one of the electrodes with an absolutely normal position of the other is affected, something that can be key when identifying ratchet syndrome. And its differentiation from the other two macrodisplacement syndromes (Arias *et al,*2012; Von-Bergen *et al.,* 2007).

- Etiologic and predisposing factors

Among the predisposing factors of twiddler syndrome probably similar to those of reel syndrome are female sex, a very wide pocket that allows free movement of the generator, obesity, childhood, elderly patients and dementia are mentioned (Nicholson *et al.,* 2003).

Manipulation of the generator by the patient conscious or unconscious is cited as one of the causes; this manipulation causes a rotation on its longitudinal axis, in the case of twiddler syndrome and on its transverse axis, in the case of reel syndrome. In addition to these external factors, recognized or not, rotational phenomena of the generator into the pocket can also occur spontaneously (Gul *et al.,* 2017). Spontaneous rotation occurs without external manipulation, and this may involve the presence of a large pocket with respect to the generator (pocket-generator discordance) or may be the subcutaneous tissue is lax such as obesity and old age. These conditions have a common factor that marks the genesis

of these syndromes: the free mobility of the generator into the pocket and the consequent rotation (Gul *et al,* 2017; Elvin&Kayrak.,2011).

External rotation origin by manipulation of the patient is recognized as the main mechanism in subjects with psychiatric or mental disorders, who morbidly or uncontrollably manipulate the pacemaker pocket region (Elvin&Kayrak.,2011).

- Brief approach to reel syndrome

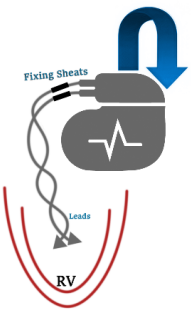
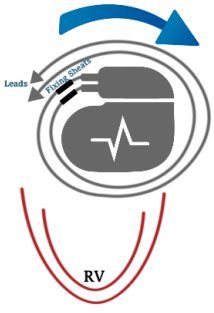
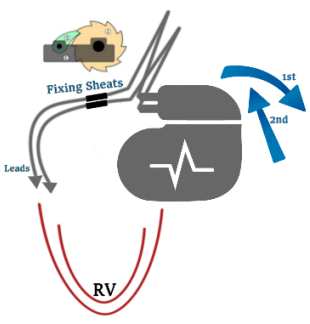
Reel syndrome is a rare cause of pacemaker and defibrillator malfunction (Carnero-Varo *et al.*, 1999). It commonly occurs within the first month after implantation, like our case report, usually does not cause damage to the stimulation electrodes, which differs from the twiddler syndrome in which generally, due to the way of the generator rotation damage to the electrodes does occur (Nicholson *et al.*, 2003).

This distinction is not purely academic; the correct recognition of kind of macrodisplacement in question has implications on the surgical treatment.

In this regard to diagnose a reel syndrome, in which there is usually no damage of the electrodes implies it is not necessary to implant a new electrode if it is verified that the electrodes are undamaged. While in twiddler syndrome should be aimed to change the electrodes once it is verified that they have been damaged, as usually occurs.

- Differences between the three macrodisplacement syndromes.

Knowledge the risk factors and mechanisms involved in macrodisplacement syndromes have a relevant importance among specialists to prevention, diagnosis and treatment these complications (Alvarez-Acosta et al., 2014). The following table shows a didactic comparison between macrodisplacement syndromes.

Aspects	Twiddler	Reel	Ratchet
Mechanism	Rotation of the generator around the longitudinal axis	Rotation of the generator around the transverse axis	Retraction and ratchet mechanism (allows the generator cross shaft to rotate in one direction but not in the opposite direction)
Consequences on electrodes	Damage may occur	No damage	No damage
Radiological aspect of the electrodes	Entanglement or knot of the leads	Reel appearance around the generator	Shrinkage without reel aspect
Occurrence moment	Within the first year	Within the first month	Within the first month
Illustrative diagram			

- Consequences of macrodisplacement syndromes.

Macrodisplacement syndromes could have varied consequences that depend on variables such as: type of device, damage of the electrodes, patient's completely dependent on pacemaker and early diagnosis, so that there may be a clinical spectrum ranging from asymptomatic patients to others with severe and life-threatening complications.

Symptoms and signs due to displacement of the stimulation electrodes towards variable sites may be: increases in the stimulation threshold, capture and sensing failures (Mahdi *et al.,* 2023), extracardiac stimulation on skeletal muscle, phrenic nerve or brachial plexus. In the case of patients with an automatic implantable cardio defibrillator (ICD)and cardiac resynchronization, the symptoms and signs may be: inappropriate therapies, worsening of heart failure due to loss of resynchronization. The most feared complication may be death due to asystole in patients totally dependent on pacing (Arias *et al,*2012; Nicholson *et al.,* 2003).

- Prevention and treatment

To prevent these rotational phenomena, some measures have been proposed at the time of implantation, such as fixing the generator to the underlying fascia or implanting it below the pectoral muscle (Von Bergen *et al.,* 2007).

Making a tight pocket can prevent free movements of the generator (Gul *et al.,* 2017) although the essential thing is to avoid discordance between the pocket and the generator, since a very small pocket can cause complications of decubitus. Several authors recommend the use of a Dacron bag around the generator as secondary prevention with good results. Radiological follow-up of predisposed patients can help make an early diagnosis and avoid major complications (Boyle *et al,* 1998; Parsonnet *et al.,* 1994).

The aforementioned constitute prevention strategies for these syndromes; however, once the complication occurs, the procedure consists of repositioning the electrodes in a correct cardiac site. Even implant new electrodes when they have been damaged because of the complication.

In ratchet syndrome, due to its production mechanism, it is essential to guarantee adequate fixation of the electrodes on their external fixation accessories (rubber cover), for which they must be tied tightly enough to prevent the electrode from slipping, but not with extreme force that could damage the sheath and silicone cover of the lead (Von Bergen *et al.,* 2007).

Another treatment option is the implantation of the leadless pacemakers when there are risk factors that cause recurrence of macrodisplacement (Montisci et al., 2024).

4. Conclusions

Knowledge about risk factors allows us to avoid lead macrodisplacement syndromes. Knowing the different production mechanisms of the three syndromes described is of great importance to finally carry out the appropriate therapy.

The reel, since it does not involve damage to the electrodes, does not require implantation of a new system, while the twiddler, since it generally involves damage to the electrodes, requires implantation of a new electrode.

The integral diagnosis allows to identify the mechanism, probable etiology, risk factors and comorbidities. This leads to the appropriate therapeutic conduct to prevent the complication from recurring.

List of abbreviations

ECG-12: 12-lead electrocardiogram

ICD: implantable cardio defibrillator

VVIR: Permanent stimulation mode in a single cardiac chamber (ventricle)

Conflict of interest

Authors not declared any conflict of interests.

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RESUMEN

Antecedentes: El desplazamiento de electrodos es una de las complicaciones relacionadas con la estimulación cardiaca. Conocer los diferentes mecanismos de producción de los macrodesplazamientos no es solo una distinción nominal. Es importante entender que los mecanismos son diferentes, por eso cada tipo de macrodesplazamiento requiere una forma diferente de terapia.

Presentación de un caso: Paciente varón de 79 años. En la exploración física, la frecuencia cardíaca era de 38 latidos por minuto. El electrocardiograma de 12 derivaciones (ECG-12) mostró un bloqueo auriculoventricular de segundo grado tipo 2:1. Se decidió la implantación de un marcapasos VVIR. En el seguimiento; la revisión del ECG-12 mostró fallo de captura ventricular sin espiga y bloqueo auriculoventricular 2:1. El estudio fluoroscópico mostró desplazamiento del electrodo. La punta distal del electrodo estaba en la zona del bolsillo del marcapasos. Se decidió inmediatamente la reintervención.

Conclusiones: El conocimiento de los factores de riesgo nos permite evitar los síndromes de macrodesplazamiento del electrodo. Conocer los diferentes mecanismos de producción de los tres síndromes descritos es de gran importancia para finalmente realizar la terapia adecuada.

Palabras clave: Macrodesplazamientos; síndrome de carrete; síndrome de Ratche; Twiddler; fallo de captura ventricular