Conclusion
This is a unique observation of the Brugada ECG in a patient with Chagas disease. It has been well documented that ajmaline can provoke a type 1 Brugada pattern in patients with Chagas disease. In addition, since this ECG pattern can be present with cardiac pathology in other diseases such as arrhythmogenic RV cardiomyopathy, it appears most likely that this characteristic ST-segment elevation is an unusual manifestation of the underlying cardiac pathology of Chagas disease.

Conflict of interest: none declared.

References

CASE REPORT

Removing the twiddling stigma: spontaneous lead retraction without patient manipulation

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After pacemaker or implantable cardioverter-defibrillator (ICD) implantation, it takes weeks for the leads to scar in place. Occasionally, newly implanted leads dislodge by retracting towards the device pocket. This phenomenon is generally called ‘Twiddler’s Syndrome,’ with the invoked mechanism being patient manipulation of the device pocket. We present a case of a 27-year-old man who had complete retraction of the atrial lead, but not the ventricular lead, after a submuscular dual-chamber ICD implantation.

It takes weeks for the leads and generator to scar in place after a pacemaker or ICD implantation. Occasionally, recently implanted leads may retract from their intracardiac position back toward, or even fully into, the device pocket. There may also be rotation of the device, which can be associated with a helical twisting of the lead upon itself. This phenomenon has been referred to as ‘Twiddler’s Syndrome,’ a term that suggests purposeful manipulation of the device by the patient. It is common for such patients to deny manipulating the device, which has led to the understanding that these patients are either unwilling to admit this behaviour or that the manipulation is subconscious.

We implanted a dual chamber ICD in a 27-year-old man with myotonic dystrophy type 1 in the setting of a PR interval of 260 ms, which is thought to be a risk factor for sudden death. Active fixation RA and RV leads were used, with both leads introduced into the venous system via a cephalic vein cut-down technique. The suture sleeves of both leads were secured to the pocket floor with two silk sutures each. Firm manual traction on each lead demonstrated good fixation within the suture sleeve, without any lead movement. A submuscular pocket was created due to a thin body habitus. Lead parameters were excellent at 6 weeks, but the atrial lead impedance abruptly increased at 2 months, associated with the loss of atrial sensing and capture. Fluoroscopy showed that the atrial lead was coiled in the device pocket, while the RV lead remained in place (Figure 1). Upon opening the submuscular pocket, the ICD generator was sutured in place with no signs of rotation, and the atrial lead was coiled in the pocket, with the ring electrode pulled back up to the firmly secured suture sleeve. A new RA lead was implanted and it remained well-positioned over time.

Implanted devices and leads are constantly exposed to friction forces, including interactions with adjacent muscle, subcutaneous tissue, and other device system components. If a lead is not fully tethered at the suture sleeve tie-down site, pocket forces may result in gradual retraction. While backward tension forces can clearly withdraw the lead out of the vasculature, forward pulsion forces are more likely to cause the flexible lead to buckle in the pocket rather than advance back into the vasculature. Consequently, when exposed to multidirectional forces over time, a ratchet-like mechanism is likely responsible for the preferential unidirectional outward migration of a moveable lead. Similarly, a non-sutured generator, when exposed to multidirectional forces, may preferentially rotate in one direction, due to its asymmetric shape. The device header typically has a rounded back end and a 90° shelf at the face on.

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which the lead connector port openings are found, making its shape not unlike the asymmetric sloped cogs in a ratchet-and-pawl mechanism that is designed to permit only unidirectional rotation.

Purposeful or subconscious device manipulation may certainly be responsible for some instances of lead retraction. In this case, however, the patient could not have preferentially manipulated only one lead in the submuscular pocket without disturbing either the other lead or the device, thereby demonstrating that leads can also fully retract without patient interference. Lead retraction has more recently been referred to as ‘Reel Syndrome,’ which removes the stigma of surreptitious or subconscious device manipulation when a patient denies ‘twiddling.’ Firm anchoring of each lead to the device pocket floor via its suture sleeve is essential to minimize the risk of lead retraction, regardless of mechanism.

Figure 1 (Left panel) Colour-inverted chest radiograph performed immediately after implantation of a left sub-pectoral dual chamber ICD, using a single coil ICD lead. The inset shows a magnified view of the active fixation right atrial lead, with a ‘J’ shape of lead slack clearly visible. (Right panel) Fluoroscopy image taken after the patient presented with lack of atrial lead sensing or capture. The atrial lead has fully retracted to the device pocket (arrow), while the right ventricular lead remains in place, with the same amount of intravascular slack as that seen on the post-implantation radiograph.

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