Case report

A 61-year-old man was referred in the intensive care unit (ICU) from an oncology department for palpitations and a wide complex tachycardia on surface electrocardiogram (ECG).

He had been diagnosed with mediastinal lymphoplasmocytic lymphoma 2 years before (2006). At first, he refused any kind of treatment. In June 2008, he considered chemotherapy. Then, he was treated with rituximab. A modified DHAP protocol consisting of oxaliplatin, cytarabine, and dexamethasone was added. The positron emission tomography scan performed as part of the baseline assessment (Figure 1, part 3) showed an abnormal uptake of the 18-fluorodeoxyglucose on the cardiac area. General tolerance of the chemotherapy was good, yet, the patient complained of palpitations 4 h after the end of the second course of treatment. Twelve-lead ECG showed a wide complex tachycardia, with left bundle branch block morphology with superior axis, atrioventricular dissociation, and fusion complexes consistent with ventricular tachycardia (VT) (Figure 1, part 1). This VT was well tolerated. It terminated spontaneously after 30 min. The ECG after the end of VT was normal. During the first 24 h of hospitalization, repetitive VT occurred (including non-sustained and sustained spontaneously terminating VT). The tolerance remained good. Morphological investigations showed that mediastinal lymphoma infiltrated the anterior wall of the right ventricle, as illustrated in Figure 1 (parts 2 and 3) and in Supplementary material online, Loop 1. The right ventricular free wall appeared thickened and infiltrated by a tumour that measured 10 × 5 cm. It was suspected that ventricular arrhythmias could be related to the tumour per se, or set off by tumour lysis after chemotherapy. Ventricular arrhythmias resumed after a loading dose of intravenous amiodarone (100 mg iv).

It was followed by an association of oral amiodarone (loading dose of 800 mg then 200 mg daily) and nadolol (40 mg daily for 2 days then 80 mg daily). During each following sequence of chemotherapy, rhythm monitoring was performed in the ICU. There was no recurrence of sustained VT. Incidence of premature ventricular contractions decreased on Holter recordings. Five months later, a second morphological evaluation was performed after chemotherapy. Complete recovery of the right ventricular function was noted, without residual tumoral infiltration (Figure 1, part 3; see Supplementary material online, Loop 2). Considering the positive evolution, amiodarone was stopped. The patient remained asymptomatic for the next 11 months.

Discussion

Cardiac involvement of mediastinum lymphoma is common and was reported in up to 75% of cases among patients with mediastinal lymphoplasmocytic lymphoma using echocardiography. However, ventricular tachycardia seems to be rare. Only a few cases have been reported in the literature. The current case illustrates the relationship between the regression of the tumoural mass under chemotherapy assessed by several imaging techniques and the resolution of the ventricular arrhythmias.

Supplementary material

Supplementary material is available at Europace online.

Conflict of interest: none declared.

References


CASE REPORT

doi:10.1093/europace/euq129
Online publish-ahead-of-print 13 May 2010

Type 1 electrocardiographic Brugada pattern in a woman with Chagas disease: a case report

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A 56-year-old woman with well-documented Chagas disease was found to have a spontaneous type 1 electrocardiographic (ECG) pattern of Brugada syndrome. It is most likely that this characteristic ST-segment elevation is an unusual manifestation of the pathological changes in Chagas disease. This ECG pattern has been found with other cardiac pathology and has been reported to be induced in patients with Chagas disease.

Case report

A 56-year-old woman with Chagas disease presented with syncopal episodes, sometimes preceded by palpitations. The diagnosis of Chagas disease was confirmed by positive immunofluorescence and haemaglutination tests to Trypanosoma cruzi. Her electrocardiogram (ECG) showed a spontaneous type 1 ECG pattern of the Brugada syndrome (BrS) (Figure 1) that was not related to anti-arrhythmic drug administration or febrile illness. This ECG pattern was not constant and sometimes reverted to a type 2
saddleback pattern. A Holter monitor showed 1376 premature ventricular contractions/24 h including ventricular triplets. She had invasive electrophysiological testing with no induction of sustained ventricular tachycardia or ventricular fibrillation. The only cardiac abnormality documented by echocardiogram and magnetic resonance imaging (MRI) was an apical left ventricular aneurysm. Implantation of an implantable cardioverter defibrillator was advised but was not approved by the insurance company.

**Discussion**

We describe the unique case of a patient with well-documented Chagas disease affecting the heart who had the typical ST-segment elevation consistent with the Brugada syndrome (BrS).\(^1\) It is most likely that the pathological changes of Chagas disease are responsible for the Brugada ECG pattern. Although the original report by Brugada indicated that there was no cardiac structural abnormality, there is now adequate documentation of cardiac fibrosis, particularly in the right ventricular (RV) outflow tract of patients with the BrS. There are morphological and clinical similarities of Chagas disease and RV cardiomyopathy. It is important to note that in 1982, before the BrS was first reported, Chiale et al.\(^2\) showed that ajmaline provoked a type 1 ECG pattern and short-coupled ventricular premature beats as in the BrS in 7 of 101 patients in the early stages of Chagas disease. Ajmaline did not provoke this ECG pattern in normal controls. This provides evidence that the type 1 Brugada ECG can be observed in patients with Chagas disease. Chiale et al. speculated that the ajmaline test could be used as a detector of myocardial damage in Chagas disease. The observation that there was no RV outflow tract abnormality by two-dimensional echocardiogram or MRI in this patient does not exclude this possibility. In a recent publication, Postema et al.\(^3\) concluded that the BrS ECG can be due to localized depolarization abnormalities and conduction delay in the RV. Hoogendijk et al.\(^4\) speculated that structural discontinuities in the RV subepicardium can cause excitation failure and activation delay by current-to-load mismatch and can cause the Brugada ECG pattern.

It is unlikely that the Brugada ECG pattern in this patient is due to a rare association of two diseases in the same individual. Genetic testing for mutations in SCN5A was not performed. Since only 15–20% of the patients with the syndrome have a genetic abnormality, negative genetic tests would not exclude the diagnosis of the BrS.

![Figure 1](image_url) Figure 1 Electrocardiogram shows maximal ST-segment elevation in right precordial leads compatible to the type 1 Brugada pattern.
Outward migration of a moveable lead. Similarly, a non-sutured generator, when exposed to multidirectional forces, may preferentially result in gradual retraction. While backward tension forces can clearly withdraw the lead out of the vasculature, forward pulsion tissue, and other device system components. If a lead is not fully tethered at the suture sleeve tie-down site, pocket forces may

Consequence of this case demonstrate that leads can spontaneously retract during normal arm movement, without any conscious or unconscious device manipulation by the patient. Leads must be firmly secured in the device pocket via their suture sleeves in order to minimize the risk of retraction, regardless of mechanism.

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,' 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. The specifics of this case demonstrate that leads can spontaneously retract during normal arm movement, without any conscious or unconscious device manipulation by the patient. Leads must be firmly secured in the device pocket via their suture sleeves in order to minimize the risk of retraction, regardless of mechanism.

Following 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. The specifics of this case demonstrate that leads can spontaneously retract during normal arm movement, without any conscious or unconscious device manipulation by the patient. Leads must be firmly secured in the device pocket via their suture sleeves in order to minimize the risk of retraction, regardless of mechanism.

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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

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References


CASE REPORT

Removing the twiddling stigma: spontaneous lead retraction without patient manipulation

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doi:10.1093/europace/euq157
Online publish-ahead-of-print 17 June 2010

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. The specifics of this case demonstrate that leads can spontaneously retract during normal arm movement, without any conscious or unconscious device manipulation by the patient. Leads must be firmly secured in the device pocket via their suture sleeves in order to minimize the risk of retraction, regardless of mechanism.

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