Junctional ectopic tachycardia and type 1 Brugada ECG in a pediatric patient: Casualty or causality?

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ABSTRACT

A 5-year-old male was admitted for tachycardia, toothache and fever. The 12-lead electrocardiogram showed a narrow QRS complex tachycardia. Following adenosine administration, the diagnosis of junctional ectopic tachycardia (JET) was established. Sinus beats showed a type 1 Brugada ECG. In the case herewith reported, JET presentation was unusual, occurring in a patient with no history of congenital heart disease or heart surgery. In the last years, Brugada Syndrome (BrS) was also correlated to various supraventricular arrhythmias. The peculiarity of the case is the first time reported association between JET and fever-induced type 1 Brugada ECG.
Introduction

Since the first report in 1992 [1], type 1 Brugada ECG has been searched for due to the potential link with life-threatening ventricular arrhythmias and sudden cardiac death. In the last years, Brugada Syndrome (BrS) was also correlated to various supraventricular arrhythmias. Giusgetetto et al. [2] demonstrated prevalence of atrial fibrillation or atrial flutter in a large cohort of patients with BrS is higher than in the general population of the same age. Recently an association between spontaneous AVNRT and drug-induced type 1 BrS was reported [3].

Case presentation

A 5-year-old male was admitted for tachycardia, toothache and fever (38 °C). Physical examination was unremarkable apart from heart rate (150 beats per minute). The 12-lead electrocardiogram (ECG) showed a narrow QRS complex tachycardia with short RP interval (about 80 ms); P waves were negative in the inferior leads and positive in lead V1, suggesting a concentric activation of the atria from bottom to top (Fig. 1). Four diagnoses were possible: (1) ectopic atrial tachycardia with long PR interval, (2) atrio-ventricular nodal reentrant tachycardia (AVNRT), (3) atrio-ventricular (AV) orthodromic reentrant tachycardia sustained by a posteroseptal accessory pathway, and (4) junctional ectopic tachycardia (JET). Vagal maneuvers (vagal-dive reflex, breath holding, Valsalva maneuver) resulted in transient slow-down of the heart rate (down to 130 bpm) without tachycardia interruption. The echocardiogram showed normal cardiac chamber size, normal ejection fraction and no heart valve disease. An intravenous bolus of adenosine (0.1 mg/kg) was administered, resulting in few sinus beats (positive P waves in lead II with normal AV conduction), followed by immediate recurrence of the arrhythmia (Fig. 2). At the beginning of tachycardia, A-V dissociation was evident (bottom row of Fig. 2), followed by resumption of retrograde atrial acti-
vation. These features established the diagnosis of JET. In the few sinus beats with normal AV conduction the QRS complex showed a type 1 Brugada ECG in lead V1 (Fig. 2, enlargement). The patient was treated with intravenous amiodarone, resulting in JET rate slowing down to 120 bpm (Fig. 3 Panel A). Two days later, propranolol was administered, with opportune pediatric dosage, restoring sinus rhythm (Fig. 3 Panel B). Antibiotic treatment was followed by fever and toothache disappearance. At discharge the ECG showed sinus rhythm with normal AV conduction, rare junctional ectopic beats and no type 1 Brugada ECG (Fig. 3 Panel C). ECG in first-degree relatives did not show Brugada Pattern. We recommended to avoid drugs reported on the website www.brugadadrugs.org and to treat promptly fever.

Discussion

Junctional ectopic tachycardia is a relatively rare arrhythmia, often occurring as a transient phenomenon following heart surgery. Congenital JET occurs in the first six months of life and is usually persistent, being associated in up to 60% of cases with heart failure [4]. In congenital JET a relatively high mortality has been reported; amiodarone and β-blockers are the most commonly used antiarrhythmic drugs for congenital or post-operative JET. Catheter ablation is a rescue therapy after failure of medical management in hemodynamically unstable patients [5,6]. In our patient, JET presentation was unusual, occurring in a 5-year-old child with no history of congenital heart disease or heart surgery. It is likely that toothache-induced adrenergic stimulus was responsible for JET.

The peculiarity of this case is the association between JET and fever-induced type 1 Brugada ECG. In the last years, BrS was correlated to various supraventricular arrhythmias [7]: atrial fibrillation /flutter and AVNRT. Genetic variants that reduce sodium channel current were found to be a possible mechanistic link between supraventricular arrhythmias and BrS, predisposing to expression of both phenotypes. [3] To the best of our knowledge, we reported, for the first time, an association between fever-induced Type 1 ECG and “idiopathic” JET. This case supports the idea that a close relationship exists between supraventricular arrhythmias and BrS.

Conflict of interest
None declared.

Funding body
None.

Ethical statement
Authors state that the research was conducted according to ethical standards.

Informed consent
The legal guardian of the little patient was asked to consider allowing Dr. Pasquale Crea to use his medical records to write a case report. The case report has been fully explained to the legal guardian and all questions have been answered. We explained to the legal guardian the objective of this manuscript, share information experienced by one patient during his clinical care that may be useful for other physicians and members of a health care team, and may be published in Cor et Vasa Journal for others to read.

The legal guardian authorized access to personal health information of the little patient.

References