

Unusual presentation of Brugada syndrome in an athlete

Ivana Nedeljkovic^{1,2}, Vojislav Giga^{1,2}, Marina Ostojic^{1,2}, Ana Djordjevic Dikic^{1,2}, Nebojsa Mujovic^{1,2}, Nenad Dikic³, Ivan Nikolic⁴, Marko Banovic^{1,2}, Tamara Stojmenović Antic^{3,4}, Olga Arsenovic Nedeljkovic^{1,5}, Ruzica Maksimovic^{1,5}, Branko Beleslin^{1,2}

¹School of Medicine, University of Belgrade, ²Cardiology Clinic, University Clinical Center of Serbia, ³Faculty of sport and management in sport, Singidunum University, Belgrade, ⁴Private practice of sports medicine "Vita Maxima", Belgrade, ⁵Center for radiology and magnetic resonance, University Clinical Center of Serbia

Abstract

Brugada syndrome is a type of arrhythmia characterized by an abnormal electrocardiogram (ECG) and an increased risk of sudden cardiac death in athletes and active recreationists. The most common sign is persistent ST elevation in ECG leads from V1-V3 with right branch block (RBBB). We presented the case of a mixed martial arts (MMA) fighter who, only at the age of 35, after increasing the intensity of training, felt fatigued, drowsiness and a drop in blood pressure to 90/60 mmHg in his recovery and at home, with the persistence of tachycardia up to 110/min. There was no syncope. The ECG showed a sinus rhythm, 62 beats/min, RBBB with suspected ST elevation in the V1 to V3 leads. The echo of the heart was completely normal as was the magnetic resonance imaging of the heart. Ergospirometry was performed and the peak oxygen consumption was 42 ml/kg/min, which indicates high functional abilities. However, the ECG showed a typical picture of type I Brugada syndrome thus confirming the diagnosis. In accordance with the recommendations, it is advisable to stop training and competition.

Key words

Brugada syndrome, sport, athletes, cardiopulmonary exercise testing

Introduction

Brugada syndrome (BrS) is an inherited disorder of the myocardial ion channels with an increased risk of ventricular fibrillation and sudden cardiac death (SCD) in people with a structurally normal heart.^{1,2,3} In addition to the electrical phenomenon, minor structural changes of the right ventricle have been described, resulting in the appearance of early repolarization.⁴ Electrocardiographic (ECG) changes typical of type I BrS occur in the form of saddle elevation of the ST segment ≥ 2 mm with a negative T-wave ≥ 1 mm in right precordial leads located in the second, third or fourth intercostal space, either spontaneously or after provocation by a sodium channels blocker^{2,4,5}. Most people with BrS remain asymptomatic during their lifetime, and the resting ECG is most impressive for the incomplete block of the right branch of the His bundle, which, in the absence of significant symptoms, is a problem for diagnosis of BrS and SCD prevention.^{1,6-8} We presented a case report of mixed martial arts (MMA) fighter with the appearance of symptoms and clear ECG changes with the image type 1 BrS only after the increased intensity of training.

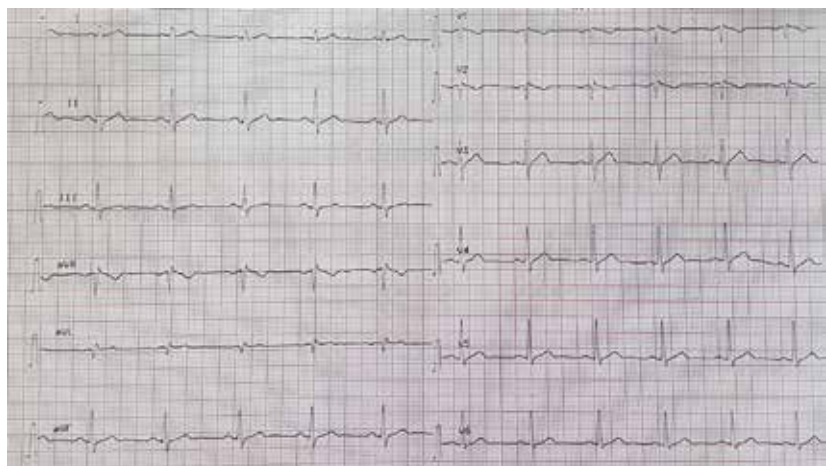


Figure 1. Electrocardiogram before cardiopulmonary exercise test

Case presentation

Dj.V, age 35, is an MMA fighter and active trainer who was until 2019 without problems during regular strenuous mixed training (endurance and aerobic training). When he started new training sessions with extremely high intensity, he felt a sudden onset of malaise and drowsiness immediately after training sessions, with a drop in arterial blood pressure to 90/80 mmHg and maintenance of sinus tachycardia with a frequency of up to 110/min.

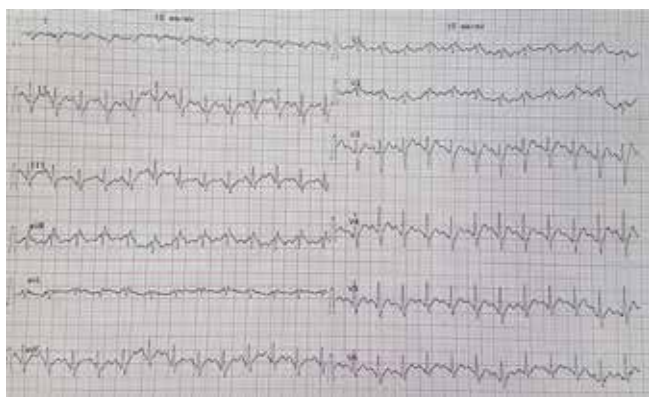
Table 1. Echocardiographic parameters

Parameters	Values
LA (mm)	35
EDD LV (mm)	56
ESD LV(mm)	33
EFLV (%)	65
IVSd (mm)	11
LVPWTs (mm)	10
E/A	0.7/0.4
E' (m/s)	0.09
E/E'	7.7
RV (mm)	24
TAPSE (mm)	29

LV - left ventricle; RV - right ventricle; LA - left atrium; EDD LV- end-diastolic dimension of left ventricle; ESD LV - end-systolic dimension of left ventricle; EF LV- ejection fraction of left ventricle; interventricular septal wall diastolic thickness; posterior wall diastolic thickness; E/A- ratio of peak velocity blood flow in early diastole and in late diastole; E'- early mitral annular velocity; E/E'- ratio of early transmitral flow velocity and early mitral annular velocity; TAPSE - tricuspid annular plane systolic excursion.

Physical examination confirmed a normal finding of the heart and lungs. At rest a 12 – lead channel electrocardiogram (ECG) recorded sinus rhythm, frequency 60 / min. with an image of the incomplete right bundle branch block (RBBB) (Figure 1). Echocardiographic examination revealed left ventricular (LV) with concentric left ventricular hypertrophy (LVH) in terms of exercise-induced cardiac remodeling (Table 1) and completely normal EF.

Considering the echocardiographic finding without signs of structural heart disease, cardiopulmonary exercise test (CPET) was performed with Schiller CS200. The test showed good functional capacity with peak oxygen consumption (peakVO₂) up to 42 mm/kg/min, which is 112% predicted, without signs of ventilatory restriction (which excludes diastolic dysfunction due to LVH) and without signs of myocardial ischemia (Table 2). However, there was a development of saddle elevation of the ST segment in the V1 and V2 leads typical of Brugada syndrome (Figure 2). ECG changes were maintained during the recovery period after the test (Figure 3).

**Figure 2.** ECG findings during cardiopulmonary exercise test presenting typical type I Brugada syndrome saddle elevation of the ST segment in the V1 and V2 leads**Table 2.** Cardiopulmonary exercise test parameters

Parameters of CPET	Values
Test duration (sec)	666
SBP start (mmHg)	120
SBP max (mmHg)	170
DBP start (mmhg)	80
DBP max (mmhg)	90
HR start (otk/min)	74
HR max (otk/min)	165
VAT VO ₂ (ml/kg/min)	16
PeakVO ₂ (ml/kg/min)	42
% estimate of VO _{2max} *	112
RER	1.2
VE/VCO ₂ slope	23

CPET- cardiopulmonary exercise test; SBP-systolic blood pressure; DBP-diastolic blood pressure; HR-heart rate; VAT -ventilatory anaerobic threshold; VO₂- oxygen uptake; Peak VO₂- peak oxygen uptake; RER- respiratory exchange ratio; VE/VCO₂ slope- ratio of minute ventilation and carbon dioxide production.

In order to completely rule out structural heart disease and active inflammatory disease, magnetic resonance (MR) imaging of the heart in standard planes was performed, which registered the preserved systolic function of the left ventricle (LV) EF 63% and right ventricular (RV) EF 66%. No zones of altered myocardial signal intensity in terms of late gadolinium enhancement (LGE) were seen on the performed native and postcontrast sequences. A homogeneous myocardial structure was registered. Also, there was a limiting width of the aorta in the bulbus up to 39 mm, with other segments of normal dimensions. Parameters of MR are shown in Table 3.

Discussion

BrS is an autosomal dominant disease with variable phenotype penetration. It is typically presented with symptoms in middle-aged males. Frequently, it can be difficult to correctly identify the ECG pattern, as it can be subtle, but as well because the pattern may occur only intermittently on ECG.

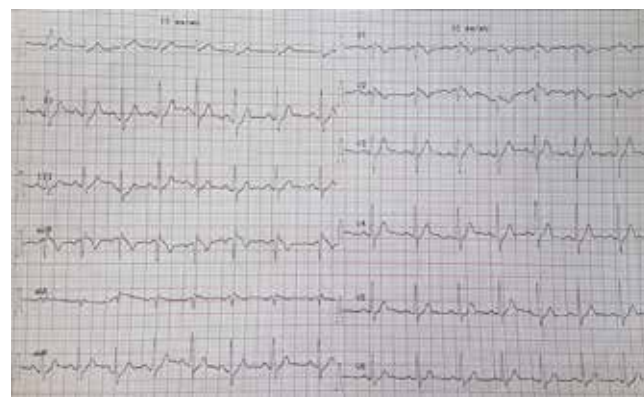
**Figure 3.** ECG changes in 3. minute of recovery after cardiopulmonary exercise test presenting typical type I Brugada syndrome saddle elevation of the ST segment in the V1 and V2 leads

Table 3. Parameters of cardiac magnetic resonance

Parameters	Values	Parameters	Values
LV		RV (mm)	24
ED (mm)	53	EF (%)	66
ES(mm)	37	EDV (ml)	152
EF (%)	63	ESV (ml)	51
EDV (ml)	195	SV (ml)	101
ESV (ml)	71	Indexed according BSA 1,1 m2	
SV (ml)	123	EDV (ml/m2)	72
Indexed according BSA 1,1 m2		ESV (ml/m2)	24
EDV (ml/m2)	92	SV (ml/m2)	48
ESV (ml/m2)	34	CI (L/min/m2)	2,53
SV (ml/m2)	58	Ascending Aorta (mm)	32
CI (L/min/m2)	3,1	LA (mm)	46x24
mass (g)	160	RA (mm)	40x39

LV - left ventricle; EF-ejection fraction; EDV- end-diastolic volume; ESV – end-systolic volume; SV- stroke volume; BSA – body surface area; CI- cardiac index; RV - right ventricle; LA - left atrium; RA – right atrium

Pelliccia et al. conducted a study comparing the ECG with standard and high leads in 491 athletes and 181 non-athletes. There were no signs of BrS on the standard ECG record, but the rSr' pattern was detected in the high leads in 76 persons in V1 and V2, of which only 2 met the BrS criteria (3%), emphasizing the importance of the high leads recording⁵.

The symptoms of BrS are also very non-specific and are related to the period of sleep or rest, so SCD in athletes can occur off the field after training, and can also be provoked by a rise in body temperature during febrile conditions or during sports or sauna^{4,7}. An unusual presentation of our case is that the symptoms appeared at the age of 35 after increasing the intensity of training. It could be hypothesized that an increased vagal response during recovery and a vagal predominance at rest may increase the susceptibility of highly trained individuals to develop arrhythmias during recovery or at rest^{4,7}.

In the case of survivors after SCD or athletes with proven syncope, ICD implantation is indicated after which return to sport is considered⁴. Symptomatic patients with ECG changes of type I BrS can compete in all sports, except in endurance sports associated with an increase in body temperature > 39C (e.g. marathon running and triathlons). Similar rules apply to the asymptomatic genotype of a positive/phenotype of a negative person and those with a latent form of BrS⁴.

Risk stratification in asymptomatic patients with spontaneous type I BrS is a major challenge. The recommendations state that evidence of deterioration in terms of clear BrS presentation during exercise or at early recovery after exercise presents a sign of increased SCD risk^{4,6}. In the case of our MMA fighter with the unmasking of BrS during the CPET and the appearance of type I Brugada, preventive measures are recommended, such as avoiding drugs that can cause (www.brugada.drugs.org) electrolyte imbalance and increase in body temperature > 39C (e.g. hot baths, saunas and steam baths, avoiding sports in hot/humid conditions or refraining from endurance training).^{4,6-8} For that cause, our MMA athlete is advised to stop strenuous training and competition.

Conclusion

In this case report, we have shown the significance of CPET in unmasking type I BrS ECG pattern in a highly trained athlete. Furthermore, for every documented Brugada pattern, expert consultation should be always obtained including the family evaluation and genetic testing.

References

1. Sieira J and Brugada P. The definition of the Brugada syndrome. *European Heart Journal* (2017) 38, 3029–3034
2. Sharma S, Drezner JA, Baggish A, et al International Recommendations for Electrocardiographic Interpretation in Athletes. *Journal of the American College of Cardiology* 2017, 69(8): 1057-75.
3. Pelliccia A, Sharma S, GatiS, et al. 2020 ESC Guidelines on sports cardiology and exercise in patients with cardiovascular disease The Task Force on sports cardiology and exercise in patients with cardiovascular disease of the European Society of Cardiology (ESC). *European Heart Journal* 2020; 00: 1-80.
4. Zorzi A, Leoni L, Di Paolo FM, et al. Differential diagnosis between early repolarization of athlete's heart and coved-type Brugada electrocardiogram. *Am J Cardiol*. 2015 Feb 15;115(4):529-32. doi: 10.1016/j.amjcard.2014.11.035.
5. Peritz DC, Chung E. Criteria for evaluating rSr' patterns due to high precordial ECG lead placement accurately confirm absence of a Brugada ECG pattern. *Journal of Electrocardiology* 2016;49: 182 – 186
6. Priori SG, Blomström-Lundqvist C, Mazzanti A, et al, 2015 ESC Guidelines for the management of patients with ventricular arrhythmias and the prevention of sudden cardiac death: The Task Force for the Management of Patients with Ventricular Arrhythmias and the Prevention of Sudden Cardiac Death of the European Society of Cardiology (ESC). Endorsed by: Association for European Paediatric and Congenital Cardiology (AEPC). *Eur Heart J*. 2015 Nov 1. 36 (41):2793-867.
7. Ackerman MJ, Zipes DP, Kovacs RJ, et al. Eligibility and Disqualification Recommendations for Competitive Athletes With Cardiovascular Abnormalities: Task Force 10: The Cardiac Channelopathies: A Scientific Statement From the American Heart Association and American College of Cardiology. *Circulation*. 2015 Dec 1. 132(22):e326-
8. Mascia G, Arbelo E, Hernandez-Ojeda J et al. Brugada Syndrome and Exercise Practice: Current Knowledge, Shortcomings and Open Questions. *Int J Sports Med*. 2017 Jul;38(8):573-581. doi: 10.1055/s-0043-107240.

Sažetak

Neobična prezentacija Brugada sindroma kod sportiste

Ivana Nedeljković^{1,2}, Vojislav Giga^{1,2}, Marina Ostojić^{1,2}, Ana Đorđević Dikić^{1,2}, Nebojša Mujović^{1,2}, , Nenad Dikić³, Ivan Nikolić⁴, Marko Banović^{1,2}, Tamara Stojmenović Antić^{3,4}, Olga Arsenović Nedeljković^{1,5}, Ružica Maksimović^{1,5}, Branko Beleslin^{1,2}

¹Medicinski fakultet, Univerzitet u Beogradu, ²Klinika za kardiologiju, Univerzitetski Klinički centar Srbije, ³Fakultet za sport i menadžment u sportu, Univerzitet Singidunum, Beograd, ⁴Privatna praksa sportske emdicine "Vita Maxima", Beograd, ⁵Centar za radiologiju i magnetnu rezonancu, Univerzitetski Klinički centar Srbije

Brugada sindrom je vrsta aritmije koja se karakteriše abnormalnim elektrokardiogramom (EKG) i povećanim rizikom od iznenadne srčane smrti kod sportista i aktivnih rekreativaca. Najčešći znak je perzistentna ST elevacija u EKG odvodima od V1-V3 sa blokom desne grane (RBBB). Predstavili smo slučaj „Mixed martial arts“ (MMA) borca koji je tek sa 35 godina, nakon povećanja intenziteta treninga osetio u odmoru i kod kuće malaksalost, pospanost i pad arterijskog pritiska do 90/ 60 mmHg uz perzistiranje tahikardije do 110/min. Nije bilo sinkopa. EKG je pokazao sinusni ritam, fr. 62 /min, RBBB sa suspektnom ST elevacijom u odvodima V1 do V3. Ehokardiogram srca je bio potpuno uredan kao i Magnetna rezonanca srca. Učinjena je ergospirometrija na kojoj je vršna potrošnja kiseonika bila 42 ml/kg/min što ukazuje na visoke funkcionalne sposobnosti. Međutim EKG je pokazao tipičnu sliku tip I Brugada sindroma čime je dijagnoza potvrđena. U skladu sa preporukama savetovan je prekid intenzivnog treniranja i takmičenja.

Ključne reči: Brugada sindrom, sport, kardiopulmonalni test fizičkim opterećenjem