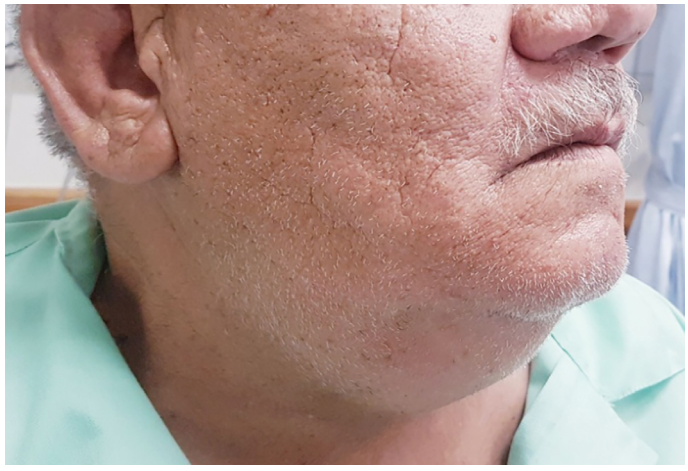

Figure 3. Partial recovery 24 hours after treatment (lateral view)

included and the possible mechanism that is proposed is edema and ductal obstruction in the salivary glands (8). Regarding our case, there are three interesting points. First, in contrast with other cases (9), in our case the kidney function test was normal. Thus, this notion that kidney failure may be involved in the pathogenesis of this adverse reaction is not plausible. Second, in our case, this adverse reaction occurred after a low dose of iodine-containing material, while in previous studies iodide mumps occurred due to excessive injection of iodine material (10). Third, although patients with iodide mumps experience painless swelling in the salivary glands, the patient in our case experienced a medium pain in the submandibular region. A study reported that iodide mumps occurred with thyroiditis (9). However, in our case the complication was isolated to the salivary glands. The routine treatment of iodide mumps is empirical with steroid, antihistamines, nonsteroidal anti-inflammatory drugs, and a combination of these medicines (11); our case received only steroid (hydrocortisone 50-mg BD intravenously) and the symptoms were resolved dramatically after 24 h. Although this unusual reaction may recur, there is no premedication to prevent this complication (12). The purpose of this case report is to make interventional cardiologists aware of this rare complication in patients who have undergone coronary angiography and have received iodine-containing contrast.

Conclusion

Iodide mumps is a rare side effect of iodine-containing contrast after coronary angioplasty that resolve after short period of time without any complication.

Informed consent: Informed consent was obtained from this patient.

References

1. Gilgen-Anner Y, Heim M, Ledermann HP, Bircher AJ. Iodide mumps after contrast media imaging: a rare adverse effect to iodine. Ann Allergy Asthma Immunol 2007; 99: 93-8.
2. Christensen J. Iodide mumps after intravascular administration of a nonionic contrast medium: case report and review of the literature. Acta Radiol 1995; 36: 82-4.
3. Azeemuddin M, Chaudhry MBH, Shahid J, Belal SZ. Non-ionic iodinated contrast-induced sialadenitis with parotid gland sparing in patient of hepatocellular carcinoma. BMJ Case Rep 2018; 2018. pii: bcr-2017-222761.
4. Egan M, Maglione PJ. Multiple reasonably tolerated percutaneous coronary interventions in a patient with iodide mumps. Ann Allergy Asthma Immunol 2015; 115: 253-4.
5. Miller J, Sussman RM. Iodide mumps after intravenous urography. N Engl J Med 1956; 255: 433-4.
6. Chuen J, Roberts N, Lovelock M, King B, Beiles B, Frydman G. "Iodide mumps" after angioplasty. Eur J Vasc Endovasc Surg 2000; 19: 217-8.
7. Chow KM, Wong KT, Szeto CC. A lady with rapid onset of swollen parotid glands. South Med J 2008; 101: 428-31.
8. Bohora S, Harikrishnan S, Tharakan J. Iodide mumps. Int J Cardiol 2008; 130: 82-3.
9. Moisey RS, McPherson S, Wright M, Orme SM. Thyroiditis and iodide mumps following an angioplasty. Nephrol Dial Transplant 2007; 22: 1250-2.
10. Jiao A, Farsad K, McVinnie DW, Jahangiri Y, Morrison JJ. Characterization of Iodide-induced Sialadenitis: Meta-analysis of the Published Case Reports in the Medical Literature. Acad Radiol 2019; pii: S1076-6332(19)30243-0.
11. Greco S, Centenaro R, Lavecchia G, Rossi F. Iodide mumps: sonographic appearance. J Clin Ultrasound 2010; 38: 438-9.
12. Lucarelli A, Perandini S, Borsato A, Strazimiri E, Montemezzi S. Iodinated contrast-induced sialadenitis: a review of the literature and sonographic findings in a clinical case. J Ultrason 2018; 18: 359-64.

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Delayed diagnosis of short QT syndrome concealed by pacemaker implant due to sick sinus syndrome

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Introduction

Patients with rare diseases unfortunately confront with late or misdiagnosis issues due to unawareness and ignorance of ordinary physicians inexperienced in related subject. Short QT

syndrome (SQTS) is a rare and highly mortal inherited channelopathy. In this case report, we present delayed diagnosis of a SQTS case and underscore red flags to avoid misdiagnosis with sick sinus syndrome (SSS).

Case Report

A 26-year-old woman suffering from brief dizziness periods and atypical chest pain was evaluated. A VVI pacemaker was implanted for SSS at the age of 8 years. Atrial fibrillation (AF) was detected when the battery was replaced; on follow-up, she experienced frequent episodes of AF with rapid ventricular response. At the age of 21 years, her pacemaker was upgraded to DDD mode following a successful radiofrequency ablation of AF. Despite amiodarone treatment, AF recurred 18 months later, and electrical cardioversion failed to restore sinus rhythm. On presentation, she was on aspirin and metoprolol, and her physical examination was unremarkable except for a pansystolic murmur at the tricuspid area. Electrocardiography (ECG) indicated AF and pure ventricular pacing (Fig. 1a). Echocardiography displayed normal left ventricular function, mildly dilated left atria, and moderate tricuspid valve regurgitation. Laboratory findings were normal including serum electrolyte levels. Pacemaker interrogation indicated a chronically elevated pacing threshold. Telecardiogram displayed retracted ventricular lead, whereas atrial lead was overlooped and bended toward the tricuspid valve. There was no evidence of significant dysrhythmia or pacemaker malfunction on prolonged Holter monitoring, but QT interval variation between paced and conducted beats was noticed (Fig. 1b). A repeat ECG revealed short QT interval (Fig. 1c) and was confirmed by sinus rhythm tracings found in past medical records (Fig. 1d). Family history was alarming as her father and grandfather had sudden cardiac deaths (SCDs). Her father was also diagnosed with SSS, and a VVI pacemaker was implanted before his SCD. Her father's available ECG tracing was consistent with AF and paced rhythm with a short QT interval more prominent in conducted beats (Fig. 1e). Therefore, a genetic testing was performed, and p.V141M_ *KCNQ1* mutation was identified confirming SQTS. Metoprolol was switched to sotalol. In addition, an implantable cardioverter defibrillator (ICD) upgrade procedure was performed involving extraction of the pacemaker leads.

Discussion

Diverse presentations of SSS involves chronotropic incompetence, episodic/persistent bradycardia, sinus pause/arrest,

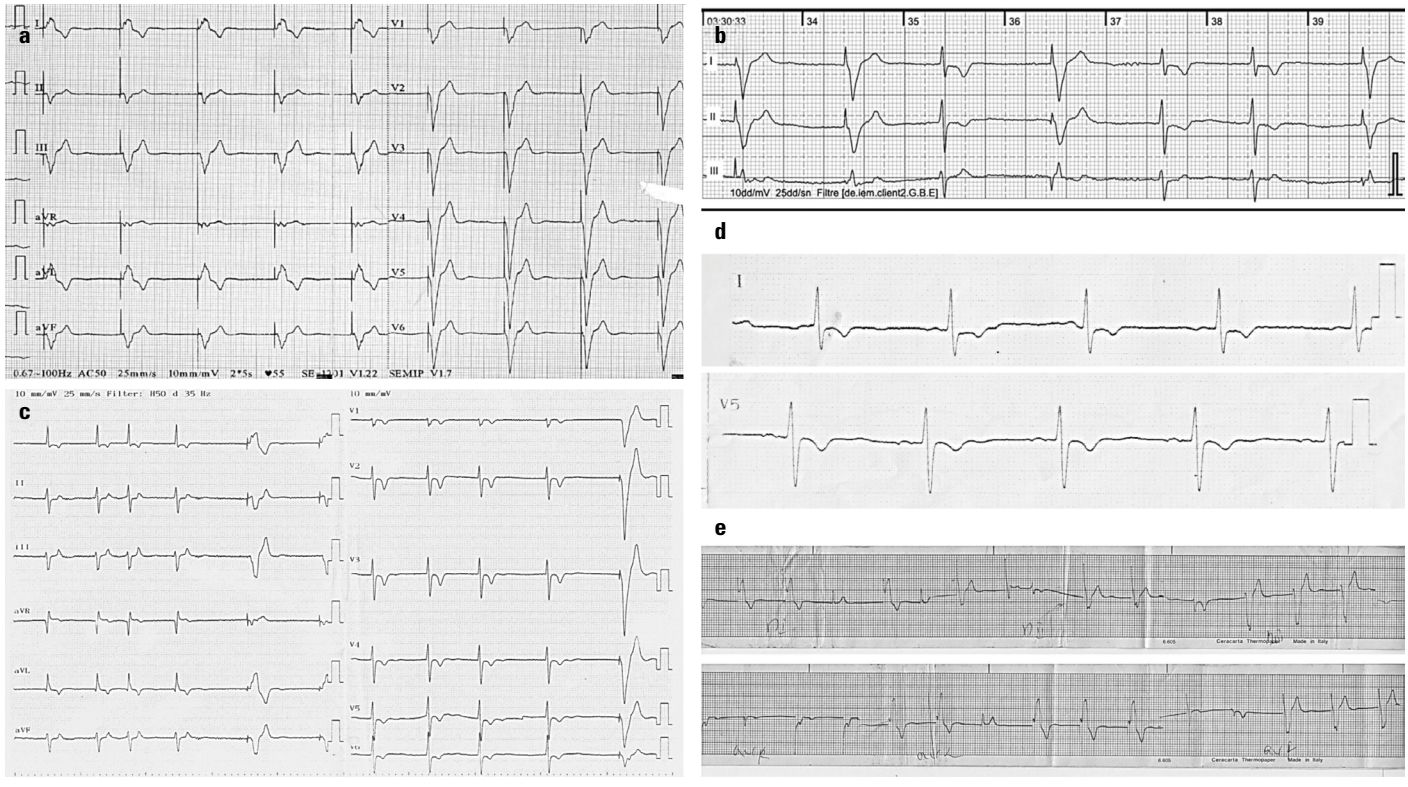

Figure 1. (a) The initial ECG captured during AF and pure ventricular pacing masks SQTS (corrected QT: 402 ms). (b) Event recording displays prominent QT variation between paced (400 ms) and conducted (320 ms) beats. (c) A repeat ECG suggests SQTS diagnosis with short QT interval (QT: 280 ms and corrected QT: 340 ms) prominent during conducted beats. (d) Sinus rhythm tracings found in past medical records confirm SQTS (corrected QT: 330 ms) in the index patient. (e) Deceased father's available ECG tracing also confirms SQTS as QT interval was short in both paced and conducted beats during AF

and occasional atrial tachyarrhythmias (1). It accounts for considerable amount of pacemaker implantations. Comprehensive clinical and ECG evaluation to assure symptom–rhythm correlation is a must before pacemaker implantation. Because SSS shares common symptoms (fatigue, palpitations, dizziness, or syncope) and components (sinus dysfunction and AF) with inherited primary arrhythmia syndromes and in some cases bradycardia might be a consequence of atrial tachyarrhythmias. SQTS is a rare disease associated with atrial and ventricular tachyarrhythmias and high risk of SCD (2). From sinus node dysfunction to AF, different aspects of SQTS may manifest in an evolutionary pattern as previously described (3), and short QT interval might be unrecognized such as the presented case. Syncope as initial presentation is not uncommon in SQTS, but generally, its association with SQTS cannot be established, resulting in delayed diagnosis (2). Thus, marked bradycardia or AF in childhood and adolescence should alert physicians to suspect SQTS (2). SQTS is diagnosed (4) if the patient's corrected QT interval is ≤340 ms. If it is ≤360 ms, one of the following additional criteria is required: (1) confirmed pathogenic mutation, (2) family history of SQTS, (3) family history of SCD before age <40, and (4) survival from a VT/VF episode in the absence of structural heart disease. In our case, short QT interval was confirmed during conducted ventricular beats, but not in paced beats. However, short QT interval in the past ECG tracings was overlooked, and she and her father were both misdiagnosed with SSS. Prolonged QT interval during ventricular pacing may obscure the diagnosis, possibly when a purely paced ECG is obtained. Because pacing induced myocardial depolarization conduction wave is slower, hence the repolarization is also longer. Currently, mutations in three potassium (KCNH2, KCNQ1, and KCNJ2) and L-type cardiac calcium channel genes have been defined in association with SQTS (2, 4). In this case a highly pathogenic p.V141M_ *KCNQ1* mutation was identified warranting the SQTS diagnosis which is particularly related with cardiomyopathic changes in the left ventricle (5). Her family history supported the diagnosis and poor outcome expectation. There is no validated risk stratification scheme for SQTS, and electrophysiological study has no added value. Theoretically, patients may benefit from antiarrhythmic drugs that prolong QT interval. Hydroquinidine or sotalol is offered in asymptomatic patients with a family history of SCD or to those who refuse ICD implantation when indicated (4). Although the beneficial effect of sotalol is lacking in certain mutations, sotalol was prescribed until the preferential choice hydroquinidine (4, 6) is obtained from abroad. The patient's dizziness complaints may be related to nonsustained ventricular tachycardia episodes which could not be captured by prolonged Holter monitoring. However, individually, pacemaker leads were extracted, and an ICD was implanted. Currently, ICD implantation is offered only to survivors of aborted cardiac arrest and patients with SQTS with documented episodes of ventricular tachycardia (4). Based on strong family history of SCD and additional ECG evidence of SQTS diagnosis in her deceased father and reported dismal prognosis in p.V141M mutations, ICD implantation was considered beneficial in such a young patient. The leads requiring revision due to their unfavorable position also influenced our decision.

Conclusion

SQTS is a highly lethal inherited cardiac channelopathy. Initial presentation may simulate SSS. When an SSS diagnosis is established, especially in children and young patients with slow heart rates and AF, the cardiologists should also keep in mind the possibility of SQTS. A careful systematic ECG analysis in pacemaker patients and comprehensive family history may avoid misdiagnosis as SSS.

Informed consent: An informed consent was obtained from the patient.

References

1. DePonti R, Marazzato J, Bagliani G, Leonelli F, Padeletti L. Sick Sinus Syndrome. Card Electrophysiol Clin 2018; 10: 183-95. [CrossRef]
2. Bjerregaard P. Diagnosis and management of short QT syndrome. Heart Rhythm 2018; 15: 1261-7. [CrossRef]
3. Righi D, Silvetti D, Drago F. Sinus bradycardia, junctional rhythm, and low-rate atrial fibrillation in short QT syndrome during 20 years of follow-up: three faces of the same genetic problem. Cardiol Young 2016; 26: 589-92. [CrossRef]
4. Priori SG, Blomström-Lundqvist C, Mazzanti A, Blom N, Borggrefe M, Camm J, et al.; ESC Scientific Document Group. 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; 36: 2793-867. [CrossRef]
5. Sarquella-Brugada G, Campuzano O, Iglesias A, Grueso J, Bradley DJ, Kerst G, et al. Short QT and atrial fibrillation: A KCNQ1 mutation-specific disease. Late follow-up in three unrelated children. HeartRhythm Case Rep 2015; 1: 193-7. [CrossRef]
6. Mazzanti A, Maragna R, Vacanti G, Kostopoulou A, Marino M, Monteforte N, et al. Hydroquinidine prevents life-threatening arrhythmic events in patients with short QT syndrome. J Am Coll Cardiol 2017; 70: 3010-5. [CrossRef]

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