Case Report

PERI AND POST-OPERATIVE MANAGEMENT OF A PATIENT WITH BRUGADA SYNDROME UNDERGOING MANDIBLE CYST REMOVAL: A CASE REPORT

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ABSTRACT

Brugada syndrome (BS) is an inherited cardiac disease that can lead to SCA (sudden cardiac arrest) in healthy young patients with structurally healthy hearts. Several treatments in common dental practice may be involved in the genesis of life-threatening arrhythmias, and local anesthetics themselves can have a potential role in eliciting the disease. Since surgical procedures can trigger the genesis of ominous arrhythmias, the setting -- of proper peri and post-operative protocols is mandatory when treating this type of patient. Anesthesiologic and cardiovascular risk must be conducted with particular care, and some procedures, such as placement of an external defibrillator along with continuous blood pressure and ECG monitoring, are needed to prevent the potential onset of arrhythmias. BS is a life-threatening condition, and despite its relatively low incidence, dentists should be aware of related risks since even simple local anesthesia may trigger a fatal arrhythmia. The aim of this case report is to describe the peri and post-operative management of a patient with BS undergoing mandible cyst removal.

KEYWORDS: *Brugada syndrome, oral surgery, cyst, dental extraction*

INTRODUCTION

Brugada syndrome (BS) was described in 1992 (1) as an inherited arrhythmic channelopathy characterized by the presence of specific ECG alterations at rest and by the occurrence of life-threatening malignant tachyarrhythmias.

Brugada syndrome (BS), also known as Nava-Martini-Thiene syndrome, was named after three Italian researchers who first described a condition that causes sudden cardiac death or ventricular tachyarrhythmias (2). Clinical

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manifestations can vary from syncopal episodes or palpitations to ventricular tachycardia/fibrillation and cardiac arrest. Still, most patients with BS are asymptomatic, and SCA may be the first manifestation. BS is an endemic disease in southeast Asia, which is often referred to as SUNDS (sudden unexplained nocturnal death syndrome), and it is considered the prevalent cause of sudden death among young people without structural cardiac anomalies. BS is thought to be responsible for 4-12 % of all SCAs and 20% of those in a structurally normal heart (3).

True incidence, due to its variable clinical manifestation, is challenging to estimate, and symptoms can occur spontaneously or be triggered by various factors such as fever, electrolyte imbalance, increased vagal stimulation, or even emotional stress. The mean age at presentation of symptoms is approximately at the fourth decade (4), and it has a tendency to affect males (5) (male-to-female ratio of 8:1). In rare cases, it can occur in childhood. It may play a potential role in sudden infant death syndrome (SIDS) (6).

The syndrome takes its name from Josep and Pedro Brugada, who, in 1992, presented several cases of patients reporting aborted sudden cardiac arrest without structural cardiac anomalies (1). However, since 1953 (7), reports of ECG patterns resembling BS are present in literature, remarking on the elusiveness and the difficulty of defining this pathology. BS is considered an autosomal-dominant transmission disease with incomplete penetrance (8). Mutations of specific genes linked to subunits of ion channels brought to an alteration of the sodium, calcium, or potassium currents in cardiac cells, producing characteristic ECG patterns and related clinical manifestations.

Loss-of-function mutation in SCN5A (a sequence related to a subunit of a sodium cardiac sodium channel) is found in up to 28% of BS patients, with more than 300 alterations reported. The mechanisms underlying arrhythmia in BS depend on the genesis of ionic disorders generated by transmural dispersion of repolarization or abnormal conduction pathway (repolarization and conduction hypothesis) occurring in the right ventricular outflow tract (RVOT). The spreading of these ionic currents through the epicardium can trigger arrhythmias and elicit the pathology (9). Diagnosis relies on identifying specific patterns in at least one of the suitable precordial leads (V1, V2, V3). There are three types of patterns, but only one (type 1) is considered diagnostic for BS; the other two (type 2 and type 3) suggest the pathology.

Even if not diagnostic, they can evolve in pattern 1 using certain classes of medications, such as sodium channel blockers (ajmaline and flecainide). Pattern type 1 is characterized by a “coved type” ST-elevation over 2 mm from the isopotential line and negative T-wave.

Type 2 and 3 (“saddleback”) show a high initial augmentation of ST-elevation over 0,5 mm followed by a convex ST segment and a positive T-wave in V2. In patients with patterns 2 and 3, provocation tests can unmask the pathology and induct a type 1 pathognomonic ECG pattern (4).

ECGs of BS patients can vary greatly, and the pathognomonic trace (pattern type1) may even disappear (10). Furthermore, different factors could act to mask the syndrome (11), highlighting its extreme variability over time. Despite numerous attempts to find a practical pharmacological approach, there is currently no available treatment that significantly reduces the number and severity of clinical manifestations, and risk stratification is an essential part of managing these patients.

Nowadays, implantable cardioverter-defibrillator (ICD) seems to be the most effective solution to reduce mortality in BS patients. However, new solutions, such as catheter ablation, in certain conditions, showed significant efficacy in reducing the arrhythmic risk (12) actively (conversely by ICD). Screening of all BS patients’ families is indicated and should involve ECG and flecainide or ajmaline test. EPS’s role in risk stratification is disputable. Some studies reported a high incidence of false positives and negatives (13), and reproducibility of results seems to be another critical issue (14, 15).

Due to the high rate of complications, ICD implantation should be avoided in asymptomatic patients (16). According to the latest consensus in 2015 on the management of BS in these patients, the presence of spontaneous or induced type 1 Brugada pattern, familiar history of SCA, and/or the inducibility of VT or VF during an EPS are factors to be considered in the decision to place an ICD (17).

Many drugs have been documented to trigger the type 1-ECG pattern; patients should be informed and aware of these “Brugada drugs” (18) to avoid the rising of arrhythmic storms. Certain medications, such as beta-blockers or calcium antagonists, should be preferably avoided in BS patients due to their action on the ST segment (19, 20).

Regarding dental practice, local anesthetics with slow dissociation properties, such as bupivacaine, levobupivacaine, and ropivacaine, should be avoided. Faster dissociation anesthetics, such as lidocaine, must be preferred (22). Furthermore, the use of epinephrine is a debated topic. Even if it is categorized as “Brugada drugs” (18), its role can be crucial for reducing the systemic administration of local anesthetics, and its use in combination with lidocaine is safe in clinical practice (23). The purpose of this article is to describe a case of cyst removal in a patient with BS a patient with BS.

CASE REPORT

A 43-year-old male patient with a radiolucent lesion in the symphysis area was referred by his general dentist to our Department of Oral Surgery, University of Chieti, Italy. The patient's familiar anamnesis was negative for SCA, and he was known to have an ECG compatible with a Brugada type 1 pattern on a flenicaide test in 2021. Since the absence of symptoms and the presence of specific ECG alterations, cardiologists decided to follow up with the patient without therapy. He underwent several surgical procedures in the past without complications. According to the American Society of Anesthesiologists (ASA), the patient was classified as an "ASA4" since a high risk of developing malignant arrhythmias was found. The treatment plan consisted of cyst removal under local anesthesia. The patient had already taken amoxicillin 1 gr of the tablets twice daily in the 3 days before surgery. Panoramic radiogram and TAC Cone Beam 3D showed radiolucency in the anterior mandible (Fig. 1-2).

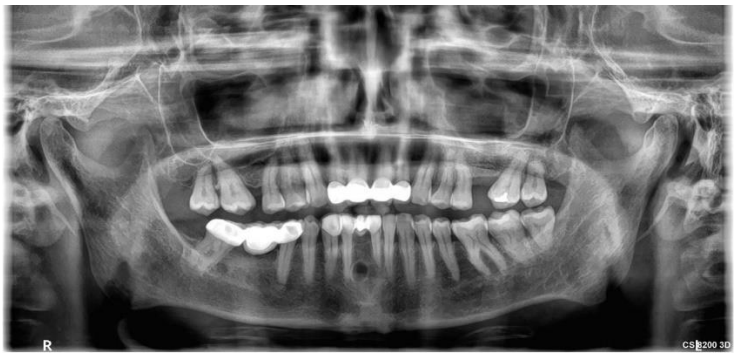

Fig. 1. *Panoramic radiogram showed radiolucency in anterior mandible*.

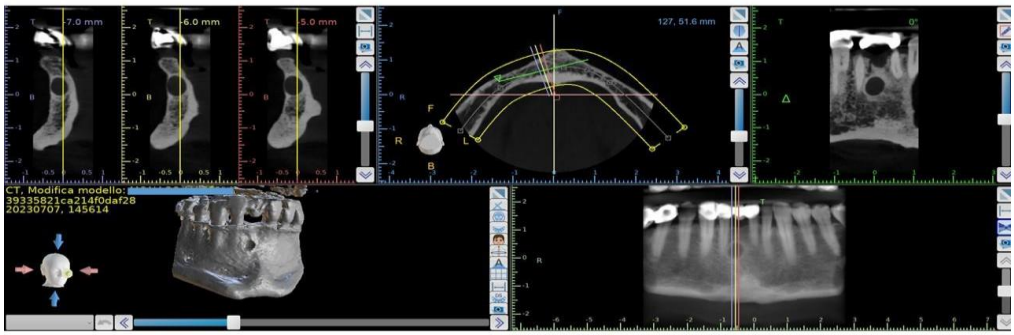

Fig. 2. *TAC Cone Beam 3D showed radiolucency in anterior mandible*.

Intraoral examination displayed swellings (in the anterior mandibular vestibule about teeth 31 and 41) that were tender to palpation. On clinical examination, teeth 31 and 32 showed good stability and were painless on horizontal and vertical percussion. The pads of an external biphasic defibrillator, 12-lead continuous ECG monitoring, and an automatic sphygmomanometer programmed to measure the patient's blood pressure every 5 minutes were placed. Isoproterenol was ready for emergency intravenous infusion (24). Block analgesia of the left and right mental nerve was implemented with block 2% mepivacaine with adrenaline (3M ESPE, Seefeld, Germany). A triangular flap (25, 26) (Fig.3) was performed, then an osteotomy was performed, and the cyst was removed (Fig. 4).

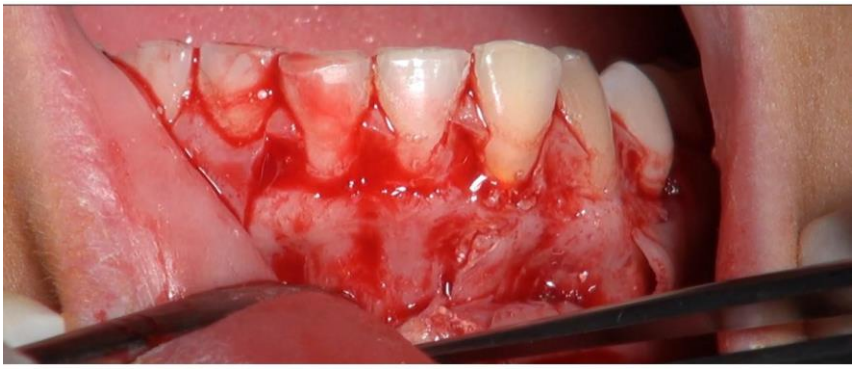

Fig. 3. *A triangular flap was performed, then an osteotomy was performed, and the cyst was removed*.

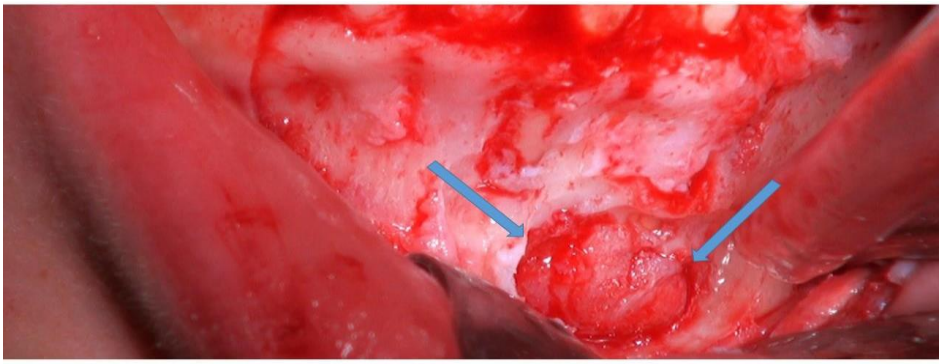

Fig. 4 *Clinical aspect of lesion*

ECG did not show any significant change. After the procedure, the patient was accompanied to a separate room where ECG monitoring continued for 2 hours; ECG did not show any significant change.

DISCUSSION

Cardiovascular arrest risk in BS patients is well described in the literature (27), and management of that risk is a fundamental part of these patients. Several articles highlight the need to avoid certain drugs commonly used in oral surgery. Regarding dental practice, local anesthetics with slow dissociation properties, such as bupivacaine, levobupivacaine, and ropivacaine, should be avoided. Bupivacaine, for instance, was shown to induce the Brugada-like electrocardiographic pattern in silent carriers of SCN5A mutations (28).

Lidocaine was shown to be safe in clinical practice (23); however, Barajas-Martinez et al. (29) reported, in rare cases of double mutation of cardiac sodium channels, a specific and pathognomonic ECG type-1 pattern even with its use. Local regional anesthesia must be performed with caution in BS patients; doses should be minimized, and active control of the patient's vital parameters should be conducted. Assisted local anesthesia, as well as the use of epinephrine, can be an excellent choice to reduce doses and risk of systemic administration of the local anesthetics. Since every increase in vagal tone can raise ST elevation, anxiety and pain must be avoided by a conscious sedation protocol. Benzodiazepines didn't seem to be associated with ECG changes (30), and their use could be crucial for stabilizing the vagal tone.

Placement of an external defibrillator, blood pressure, and ECG monitoring must be carried out throughout the treatment, and, in the absence of arrhythmias, isoproterenol (a beta-receptor agonist) can be used to reduce ST-segment elevation (30). In patients with ICD, according to the cardiologist, the device should be switched off to prevent inappropriate shocks due to monopolar surgical diathermy (22). ECG monitoring should be maintained during the first 24 hours after surgery (22).

BS is a life-threatening condition and requires special measures. Communication between the dentist and cardiologist is necessary to assist those patients properly. Particular attention must be given to anesthetic management. The cardiologist and anesthesiologist must accurately choose the type of anesthesia based on the patient's general assessment and risk. The dentist must analyze the patient's medical history to detect a family history of sudden death or ischemic disease so that further investigations can be performed before treatment.

Due to the lack of large trials, there are no clear recommendations for general or regional anesthesia in BS patients. In conclusion, managing BS requires special vigilance, avoiding factors and situations that potentially trigger arrhythmias, and setting up proper peri and post-operative protocols.

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