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CASE REPORT

ADVANCED

CLINICAL CASE

Hypoglossal Nerve Stimulator-Induced Neurapraxia Following Electrical Cardioversion for Atrial Fibrillation

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ABSTRACT

An increasing proportion of patients with atrial fibrillation are undergoing implantation with hypoglossal nerve stimulators for the treatment of obstructive sleep apnea. We present a case of hypoglossal nerve stimulator–associated neurapraxia following electrical cardioversion of atrial fibrillation. (**Level of Difficulty: Advanced.**) (J Am Coll Cardiol Case Rep 2021;3:1128–31) © 2021 Published by Elsevier on behalf of the American College of Cardiology Foundation. This is an open access article under the CC BY-NC-ND license (http://creativecommons.org/licenses/by-nc-nd/4.0/).

HISTORY OF PRESENTATION

A 78-year-old man with obstructive sleep apnea (OSA) treated with a hypoglossal nerve stimulator (HNS) presented for management of symptomatic, persistent atrial fibrillation (AF). He initially received a diagnosis of AF in 2016 and was treated with a rate-control strategy. Over time he was noted to develop progressive dyspnea, and the decision was made to pursue a rhythm control strategy with dofetilide. His baseline examination findings on presentation were as follows: blood pressure 134/95 mm Hg; heart rate 78 beats/min; afebrile; pulse oximetry 96% on ambient air; obese male weighing 137.9 kg in no distress; neck supple without jugular venous distention; no murmurs, rubs, or gallops with equal, but irregular pulses; lungs symmetrical and clear; skin without rashes; neurologically nonfocal grossly; awake, alert, and oriented. The patient was admitted for drug loading and underwent electrical cardioversion (ECV) with a single 200-J shock delivered from anterior to posterior with restoration of sinus rhythm. On subsequent activation of his HNS, the patient experienced a painful sublingual "shock"-like sensation with greater than anticipated tongue deviation to the left. The HNS was deactivated, with amelioration of his symptoms; however, the same symptoms returned on immediate attempts at reactivation, thus precluding further use of the HNS.

LEARNING OBJECTIVES

- To recognize the increasingly common cohort of patients with OSA treated with an HNS who have AF and are in need of ECV.
- To understand the risk of injury as well as both temporary and irreparable damage to the HNS from ECV.
- To distinguish the protective circuitry differences between an HNS and a pacemaker or defibrillator with respect to ECV.
- To understand the outlined approach to avoid patient injury and/or HNS-related compromise whenever a patient with AF and an HNS is undergoing ECV.

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The authors attest they are in compliance with human studies committees and animal welfare regulations of the authors' institutions and Food and Drug Administration guidelines, including patient consent where appropriate. For more information, visit the Author Center.

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PAST MEDICAL HISTORY

In addition to longstanding, persistent AF and OSA treated with an HNS, the patient had a history of nonobstructive coronary artery disease, hypertension, hyperlipidemia, and obesity. Of note, OSA treatment with continuous positive airway pressure (CPAP) therapy had failed because of claustrophobia. He underwent implantation with an HNS in 2018.

DIFFERENTIAL DIAGNOSIS

The chief suspicion was an electrical malfunction of the HNS hardware stemming from the ECV just hours earlier, given its proximity to symptom onset. Alternatively, HNS electrode-related compromise or dislodgment and direct neural injury at the tissue-lead interface were considered.

INVESTIGATIONS

Direct examination of the patient both at rest (**Figure 1A**) and with HNS activation (**Figure 1B**) established the device-associated complication, characterized by exaggerated and painful leftward tongue deviation. Lateral neck and chest radiography (**Figure 2**) documented relative anatomical stability and device hardware integrity compared with the implant studies.

MANAGEMENT (MEDICAL OR INTERVENTIONS)

The HNS remained deactivated until follow-up with the patient's implanting otorhinolaryngologist the day after hospital discharge. Reactivation 48 hours post-ECV yielded less severe symptoms, although mild sublingual tenderness remained during stimulation at his baseline settings of 1.5 V at the default electrode configuration, pulse width, and rate. Interrogation of the device demonstrated stable functional parameters characterized by a sensation threshold (stimulation appreciated by patient) of 0.7 V and a functional threshold (tongue motion observed) of 1.0 V. Given the lingering symptoms, the outputs were reduced to maintain adequate function while avoiding the "shock"-like sensation, settling on 0.8 V.

DISCUSSION

There is a strong association between AF and OSA, with the incidence of each on the rise (1,2). Although CPAP therapy remains the most frequent treatment for OSA, electrical stimulation of the hypoglossal nerve has been demonstrated to be an effective alternate therapy (3). Consequently, HNS implantation has become increasingly common, and the prevalence of these devices in patients with AF has increased (4). In this case report, we present an example where ECV, a central component of AF management, resulted in a complication associated with a surgically implanted HNS. As the intersection between patients with AF and HNS for OSA management continues to grow, many clinicians will find themselves in a similar clinical scenario.

ABBREVIATIONS AND ACRONYMS

CPAP = continuous positive airway pressure

ECV = electrical cardioversion

HNS = hypoglossal nerve stimulator

OSA = obstructive sleep apnea

Failure of an HNS following ECV is a rare occurrence. A previous retrospective review of 201 patients by Vasconcellos et al (5) reported 4 instances of total device failure following repeated cardioversions for AF or direct cardioversion over the device. The devices in this series represented an older HNS technology that did not have electrical shielding and required surgical device replacement if damaged during ECV (6). Newer HNS models have protective circuitry such as is found in pacemakers and implantable cardioverter-defibrillators, namely a Zener diode. A Zener diode is distinct from other diodes in that it allows current to flow in reverse (away from the device) if the voltage exceeds a certain threshold, thereby protecting devices during ECV (7).

The case presented here differs from the previous case series because it is the first report of a device-associated complication following ECV with the newer HNS technology. Further, the mechanism of compromise was not total device failure, as in the earlier series, but was instead determined to be a form of postcardioversion neurapraxia on case review with the manufacturer's engineering staff (Inspire Medical Systems, Inc.). The HNS programming and hardware remained unaffected at subsequent device interrogation. The mechanism of neurapraxia in this case is uncertain, but it is likely related to the introduction of high voltage during the ECV causing electrical current to travel along the lead body (region of lower impedance) to the electrode-tissue interface, where a higher current density resulted in direct nerve injury. Return of noxious sensory symptoms and exaggerated motor function with device reactivation had been hypothesized to emanate from lingering residual charge on this electrode, although this remains not well understood. Hypoglossal neurapraxia stemming from ECV is an uncommon but reversible event with estimated resolution within days to weeks of the insult (3). In addition, although most of the earlier reports of HNS failure occurred in the setting of inappropriate pad placement or repeated shocks, the device-associated complication presented here occurred after a single cardioversion performed remote to the nerve stimulator.

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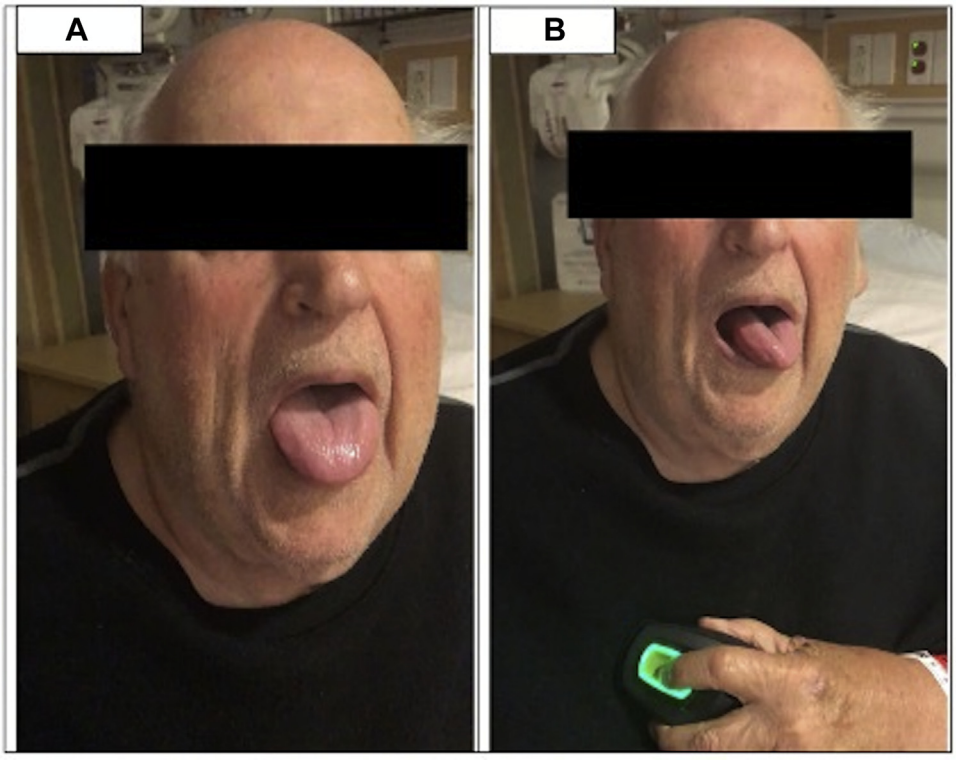
FIGURE 1 Hypoglossal Nerve Stimulator Complication After Electrical Cardioversion



(A) The patient at rest without activation of the hypoglossal nerve stimulator. **(B)** Exaggerated and painful leftward tongue deviation with hypoglossal nerve stimulator activation after electrical cardioversion.

HNS device malfunction and complications can be minimized at the time of ECV by using the following approaches (6): 1) use biphasic waveforms and minimize energy delivered; 2) position the paddles or defibrillation pads as far from the implanted HNS components as possible; and 3) after cardioversion, test or activate the HNS device to confirm that the patient can feel tongue stimulation for several seconds. If the patient cannot feel stimulation when therapy is turned on, then the patient should discontinue therapy until the device is evaluated by their otolaryngologist.

FOLLOW-UP

Post-discharge evaluation of the patient's HNS required a nonsurgical computer-driven electronic adjustment of the voltage output to achieve an appropriate and comfortable therapeutic effect with sublingual muscle recruitment. These settings were adjusted back to prehospital programming following complete symptom resolution in the week(s) that followed without ostensible irreversible damage discovered and with concurrent patient reports of restful sleep endorsed.

CONCLUSIONS

When encountering patients with AF requiring ECV, it is important to note the presence of an HNS so that steps may be taken to avoid potential device-related complications or total device failure.

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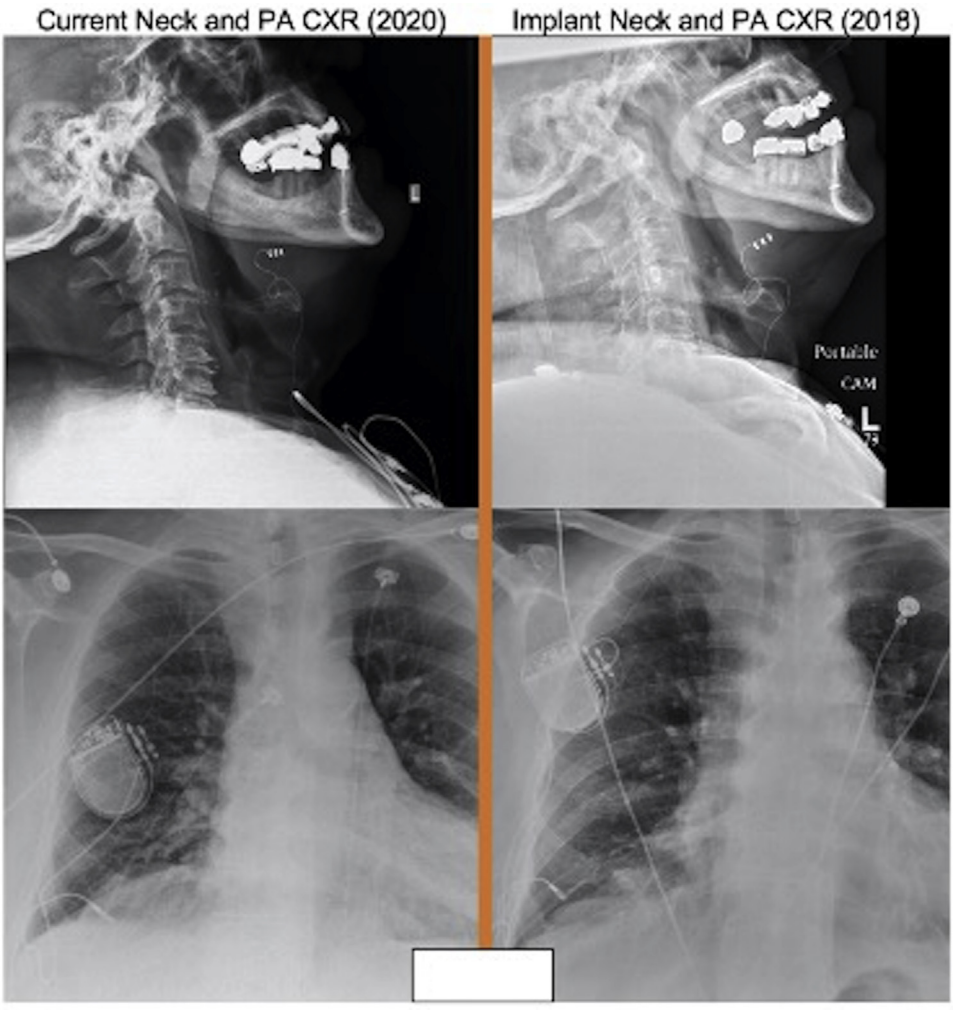
FIGURE 2 **Hypoglossal Nerve Stimulator Imaging Before and After Development of Complication**



(Top) Lateral neck and **(bottom)** posteroanterior view chest radiographs (CXR) from **(left top and bottom)** the time of hypoglossal nerve stimulator complication discovery compared with **(right top and bottom)** immediately after hypoglossal nerve stimulator implantation, demonstrating no meaningful changes. L = left.

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