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SNI: Epilepsy

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Case Report

Trigeminal paresthesia secondary to responsive neurostimulation (RNS) lead migration

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Received : 12 July 2021
Accepted : 14 October 2021
Published : 23 November 2021

DOI
10.25259/SNI_692_2021

Quick Response Code:

ABSTRACT

Background: The responsive neurostimulation system (RNS) is used in patients with drug-resistant epilepsy who are not candidates for surgical resection of a seizure focus. As a relatively new therapy option, the adverse effects of long-term implantation are still being clarified. We present a series of two patients who presented with similar symptoms which were attributable to migration of the intracranially implanted subdural leads.

Case Description: Two patients who had subdural RNS lead implantation presented with symptoms of paroxysmal unilateral facial pain which were thought to be related to the stimulation of the trigeminal nerve secondary to RNS lead migration. Adjustment of the stimulation parameters improved the symptoms in both patients.

Conclusion: Chronically implanted subdural RNS leads can migrate over time stimulating nerves in the intracranial space. Strategies to avoid and overcome the complication are discussed.

Keywords: Lead migration, Responsive neuro-stimulation, Stimulation triggered symptoms, Trigeminal nerve

INTRODUCTION

The Responsive Neurostimulation (RNS) system (Neuropace RNS® System, Mountain View, CA, USA) is a therapy for patients with drug-resistant epilepsy who are not candidates for surgical resection of a seizure focus.[1] The system involves long-term implantation of subdural or depth lead connected to a neurostimulator that detects incipient epileptic activity and delivers stimulation to control seizure spread. The intra-cranial leads are implanted as close as possible to the epileptic focus or foci to optimize detection and therapeutic stimulation ability. As with other brain-machine interface systems, RNS systems are subject to hardware-related complications.

CASE REPORT

We present two patients with symptoms of paroxysmal facial sensation ipsilateral to subtemporal RNS strip leads months after uneventful implantation. The patients did not report any sensory symptoms in the immediate post-implant period; however, they both presented independently complaining of episodes of unilateral facial sensation months later. In both patients, the symptoms developed relatively rapidly over a few weeks. Patient 1 is a 39-year-old male who developed a shock-like sensation in the right lower part of the face nearly 1.5 years after implantation.

Patient 2 is a 33-year-old male who reported tingling in the right upper part of the face approximately 9 months after implantation.

We did not appreciate any objective facial sensory abnormalities on examination in either patient. The explanation of the new symptom was not apparent initially, as the trigeminal nerve and its branches that provide facial sensation are rarely injured during this type of procedure. Furthermore, the paroxysmal nature of the reported symptoms raised the suspicion that they were related to stimulation delivery. Around the time of symptom onset, we measured impedance from the RNS contacts, and it appeared to be increased in the subtemporal leads. Impedance measurements in the other leads (hippocampal) were not altered. As lead impedance can be elevated with loss of contact with the underlying brain, we hypothesized that migration of the subtemporal RNS strips toward the intracranial portion of the trigeminal nerve might be responsible for the patients' symptoms by electrical stimulation of the nerve. There was no apparent involvement of the jaw musculature or facial muscles to suggest trigeminal motor or facial involvement.

Stimulation at the same current as the stimulation current reproduced the symptoms in the patients. Turning off the subtemporal strip caused resolution of the symptoms. We analyzed medical records and neuroimaging of the two patients. Structural brain magnetic resonance images (MRI) were co-registered with computed tomography (CT) scans to compare RNS lead locations between the immediate post-implant period and the time of symptomatic presentation. Imaging analysis of co-registered MRI-CT scans revealed anterior migration of the RNS lead tip in both cases [Figure 1]. We decided to stop the stimulation of the involved contacts to alleviate the sensory symptoms and also to test our hypothesis. Cessation of stimulation in the implicated contacts led to complete resolution of the symptoms. The contacts were turned off during subsequent treatment without an adverse seizure outcome in these patients with presumed mesial temporal seizure onsets.

DISCUSSION

We describe a unique long-term complication of RNS subdural strip implantation in the sub-temporal location. Migration of the lead can lead to stimulation of the trigeminal nerve/branches, potentially causing paroxysmal ipsilateral facial sensations or pain. Another potential explanation of the sensory symptoms can be nerve injury during lead implantation, for example, due to direct and indirect trauma related to retraction, cautery, and suction. Although uncommon, trigeminal neuropathic pain has been reported as a complication of anterior temporal lobe resection.[2] However, we perceived that this possibility is less likely since the symptoms did not start immediately or soon after the surgery and due to the periodic nature of the sensations. The fact that both patients underwent similar procedures and developed similar sensory symptoms ipsilateral to the site of lead implantation months following the intervention makes the possibility of stimulation-related complications more likely. Moreover, the co-registered brain images verified lead migration, thus corroborating our hypothesis. The original location of the leads is flush with the petrous ridge [Figure 1]. Anterior movement presumably moves the lead closer toward the anterior end of the Meckel's cave and toward the branches of the trigeminal nerve, particularly the mandibular nerve (V3) at the foramen ovale and the maxillary nerve (V2) at the foramen rotundum [Figure 2]. Patient 1 had sensations in the lower part of the face likely related to mandibular nerve stimulation and patient 2 had sensations in the upper part of the face likely related to maxillary nerve stimulation.

The term stimulation-triggered signs/symptoms (STS) has been proposed to describe uncomfortable or noticeable motor or sensory effects of stimulation in patients with RNS. In a single institution case series, STS were seen in approximately 10% (six out of 58) of RNS implants.[3] Notably, three out of six patients in this series had electrical sensation/pain in the face, all of whom had subtemporal strips ipsilateral to symptoms. Symptoms were resolved by changing stimulation parameters or turning off the involved contact. The cause for facial sensations was determined to be due to nerve stimulation, although a lead migration over time was not documented as in our case. Other STS previously reported include photopsia, muscle twitch, and dizziness.[1,3] Lead stimulation without muscle paralytics in the operating room has been suggested for early detection of motoric STS. However, sensory STS are more insidious and cannot be detected at implantation time. Management options include optimization of stimulation parameters as we did in our patients but may also involve lead repositioning if there is a concomitant worsening of seizure control. Specific optimization procedures proposed include reduction of stimulation frequency or pulse width, reduction of the current intensity, alteration of the stimulation pathway, or removal of the involved contact from stimulation.[3] Strategies for preventing such complications include anchoring subdural leads by suturing them to an adjacent dural edge, which may prevent or limit lead migration. Using shorter leads can also help limit this complication. Given that the RNS strip leads have contacts only on one side, care needs to be taken to ensure that the stimulating side is adjacent to the brain and the non-stimulating side is turned away. While it is not distinguishable whether the leads were inverted in our case, this is unlikely as the symptoms were not present at implant time and developed over time.

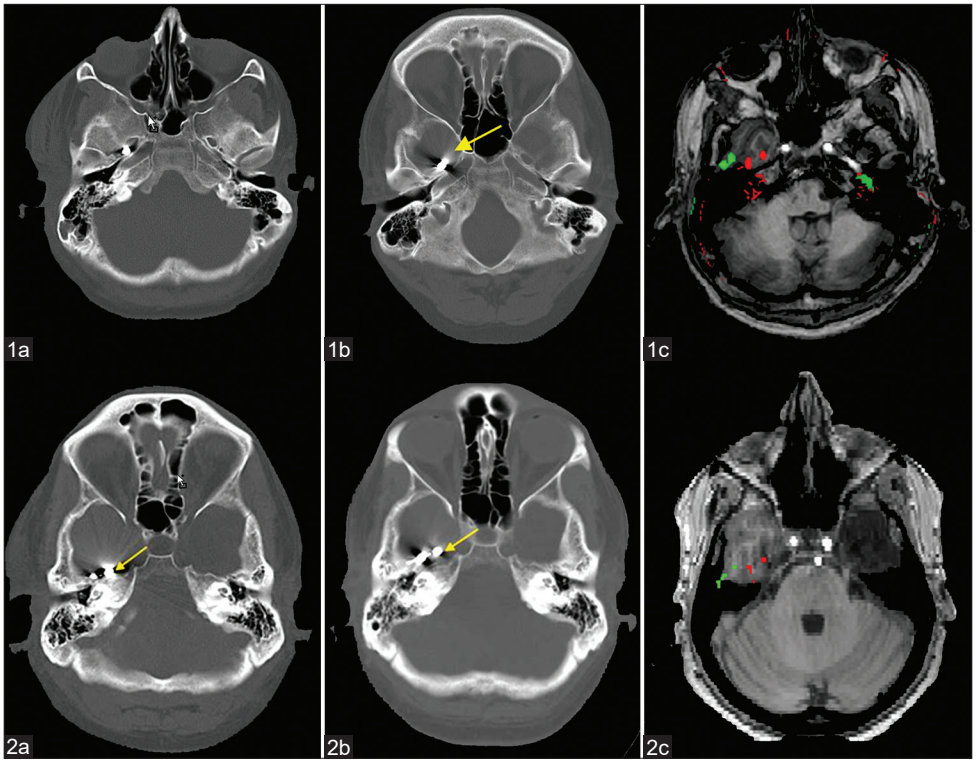

Figure 1: Images for patients 1 and 2 show the sub-temporal lead adjacent to the bone immediately after implantation (1a, 2a); repeat images reveal displacement of the same lead over time (1b, 2b). Images 1c and 2c show the relative position of the lead at implantation (red) and at presentation (green).

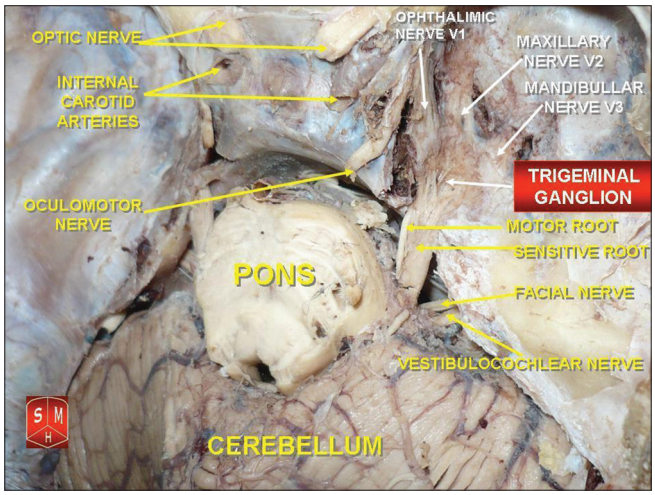

Figure 2: Representative image showing the anatomy of the middle cranial fossa and brainstem with relations between the petrous ridge, trigeminal ganglion, mandibular nerve (V3) at foramen ovale and maxillary nerve (V2) at foramen rotundum. Image credit: Anatomist 90. licensed under the Creative Commons Attribution-Share Alike 3.0 Trigeminal ganglion 2011. https://commons.wikimedia.org/wiki/File: Trigeminal_ganglion.jpg (accessed September 8, 2021).

Electrical stimulation of the sensate meninges is likely to cause sensory symptoms while stimulation of the insensate brain is unlikely to cause symptoms. It is plausible that volume conduction of electrical current to the nearby nerves led to the development of symptoms as the leads migrated closer to them. The previously described case series did not report lead inversion as a cause of STS.[3] It is interesting that our patients did not report a worsening of seizures after the involved contacts were turned off. This may be due to the beneficial effect of the uninvolved subtemporal strip contacts that continued to be stimulated, as well as the potentially greater benefit from continued hippocampal depth stimulation in these patients with presumed mesial temporal epilepsy.

CONCLUSION

Facial sensations may be associated with stimulation of subtemporal strips using the RNS system. Newly elevated contact impedances may suggest lead migration, although other reasons such as gliosis can also cause progressively elevated RNS lead impedance.[4] Careful imaging analysis can help confirm RNS lead migration. Management strategies include adjusting different stimulation parameters or turning off specific contacts. Lead re-positioning can be considered if seizure control worsens concomitant to symptom onset.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Dr. Haneef's work is funded by the Hogg foundation. Dr. Sheth is a consultant for Boston Scientific, Abbott, Neuropace, and Zimmer Biomet. Dr. Krishnan and Dr. Karimov reports no disclosures.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Haneef Z, Karimov A, Krishnan V, Sheth SA. Trigeminal paresthesia secondary to responsive neurostimulation (RNS) lead migration. Surg Neurol Int 2021;12:577.