



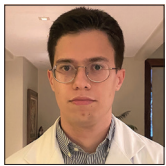
Case Report

An unusual case of lead migration in occipital nerve stimulation: A case report and literature review

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ABSTRACT

Background: Lead migration is a complication associated with occipital nerve stimulation (ONS). We present a rare case in which fibrosis in the stress relief loop caused lead migration in the treatment of occipital neuralgia.

Case Description: A 30-year-old woman with a 5-year history of refractory occipital neuralgia, who had been under ONS therapy for 2 months, presented with a sudden onset of typical occipital neuralgia pain associated with cervical muscles spasms and myoclonus. A skull radiography showed lead migration. The patient underwent surgery for lead repositioning. During surgery, we identified extensive fibrosis throughout the stress relief loop that produced several constriction points. The fibrosis in the stress relief loop increased tension on the lead during head-and-neck movement, causing progressive migration of the lead.

Conclusion: Although lead migration is a common complication of ONS, its association with fibrosis in the stress relief loop has not, to the best of our knowledge, been reported before. Lead migration can directly affect treatment outcome and it is, therefore, important to fully understand the possible mechanisms that can cause it and how to promptly manage them.

Keywords: Lead migration, Neuromodulation, Occipital nerve stimulation, Occipital neuralgia

INTRODUCTION

Occipital neuralgia is described as a paroxysmal shooting or stabbing unilateral pain in the greater, lesser, or third occipital nerve dermatome. Initial treatment includes occipital nerve block and pharmacological therapy. In the management of refractory occipital neuralgia, radiofrequency rhizotomy and occipital nerve stimulation (ONS) are therapeutic options, being ONS a more invasive procedure.^[6,13]

ONS can be used in the treatment of neuropathic pain when conservative or less invasive therapies have failed. It has been used in the treatment of occipital neuralgia, persistent chronic migraine,

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cluster headache, cervicogenic headache, and other painful disorders.^[12,16,35] Lead migration, infection, lead fracture, skin erosion, and lead disconnection are complications associated with ONS.^[9]

In this article, we report a case of a patient diagnosed with refractory occipital neuralgia that undergoes surgery for ONS and develops lead migration 2 months after surgery. An ONS system includes a lead, an internal pulse generator (IPG), and the lead extension. The lead extension connects the lead, in the occipital region, to the IPG, in the infraclavicular region. Since this is a pathway with high mobility, we produce a stress relief loop with the lead extension so that the lead does not migrate due to higher tension during flexion or extension of the head.

During surgery for lead repositioning, we observed that fibrosis throughout the stress relief loop was the cause for migration. The association of fibrosis in the stress relief loop and lead migration is a rare complication of ONS and has not, to the best of our knowledge, been reported before. We also conduct a literature review of all reported cases of lead migration in ONS in the past 10 years and discuss possible mechanisms that might cause lead migration. Lead migration directly affects treatment outcome and pain management and, hence, it is essential to understand the mechanisms that cause it, how to properly and promptly manage it, and how to prevent its recurrence.

CASE REPORT

A 30-year-old woman with a 5-year history of refractory occipital neuralgia, who had been under ONS therapy for 2 months, presented with a sudden onset of severe unilateral pain, originating in the right upper nuchal region and spreading to the occipital and parietal regions. The pain was described as stabbing, sharp, lancinating, and occurred in paroxysms. The patient also described spasms and myoclonus that occurred in the right trapezius, scalene, and supraspinatus muscles, a new symptomatic component that had not accompanied her typical occipital neuralgia pain before.

The patient had a history of microvascular decompression surgery 6 years before as a treatment for trigeminal neuralgia, with full remission of symptoms. She developed cerebral venous sinus thrombosis and mastoiditis as immediate postoperative complications, but they were appropriately and successfully managed.

Due to the failure of medical therapy for occipital neuralgia (pregabalin and duloxetine), the patient had been submitted to three percutaneous radiofrequency rhizotomies (1, 3, and 4 years ago) which provided immediate symptomatic relief and yielded significant pain control, but for a limited time. She had also received two occipital nerve blocks through local anesthesia, but with poor effect in long-term pain management.

The pain had become progressively more severe, more frequent and associated with nausea. As a result of the unresponsiveness to less invasive procedures, the patient underwent ONS 2 months ago.

ONS was employed through surgery. The patient was placed in prone position under anesthesia. A right paramedian incision was performed in the level of C1 vertebra and a pathway was carefully dissected over the muscle fascia, toward the mastoid region. Once the correct position was asserted through fluoroscopy, a lead containing 16 electrode contacts was inserted and anchored over the muscle fascia. The IPG was placed in the infraclavicular region, in a subcutaneous pocket, and connected to the lead extension, which was inserted through subcutaneous tunneling. A stress relief loop was produced with the lead cable extension in the subcutaneous pocket, alongside the IPG, forming a circular coil to minimize tension on the lead [Figure 1]. The technique provided adequate pain management after optimization of the IPG settings.

Two months after ONS were implemented, the patient suddenly started with the symptoms and manifestations described. When the ONS system was turned off, the spasms and myoclonus ceased. A skull radiography showed lead migration [Figure 2].

The patient underwent surgery for lead repositioning. During surgery, extensive fibrosis was identified throughout the lead extension loop, which was deemed as cause for the lead migration. The fibrosis produced numerous points of

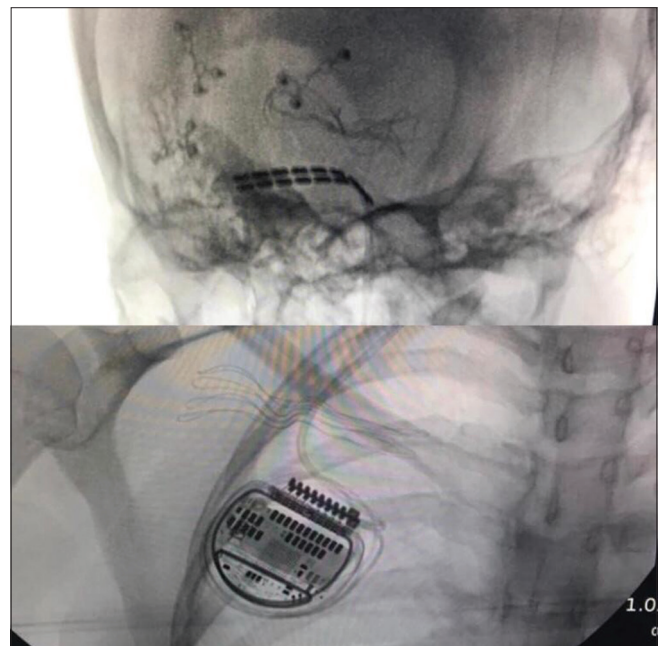


Figure 1: Lead and internal pulse generator correctly located after surgery for occipital nerve stimulation.

constriction along the stress relief loop, which provided tension over the lead during head-and-neck movement and caused its migration.

We conducted thorough dissection of the fibrotic tissue, releasing the stress relief loop, and removing the IPG from the infraclavicular subcutaneous pocket [Figure 3]. A low cervical incision was made over the dislocated lead position guided by fluoroscopy and new tunneling was performed due to important fibrosis in the subcutaneous that restrained the lead from being removed. The lead and cables were then removed through the infraclavicular incision. The lead was repositioned through the former occipital incision and the system was reinserted as described previously [Figure 4].

After lead repositioning, the patient did not experience any muscle spasms or myoclonus. The pain intensity significantly decreased and the IPG settings are still being managed to optimize pain control.



Figure 2: Skull radiography showing lead migration.

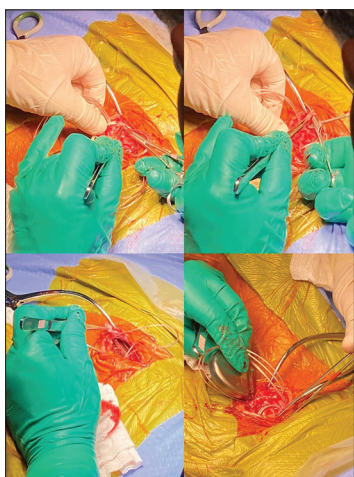


Figure 3: Surgery for lead repositioning. Dissection of the fibrosis constraining the stress relief loop can be observed.

DISCUSSION

In this case report, extensive fibrosis constricted the stress relief loop. Therefore, the loop was not able to relief tension during movement of the head and neck, which resulted in lead tension, traction, and migration. The most common complications associated with ONS are lead migration, skin erosion, infection, lead fracture, and lead disconnection.^[9]

It is hard to properly assess the incidence of lead migration in ONS since the studies that report it are heterogeneous, describe few cases, and present extremely variable results [Table 1].^[2,3,7,11,14,15,17-22,24,28-30,32,34,41] In our review, we identified five randomized clinical trials which reported lead migration.^[1,8,23,31,33] These trials have reported a migration incidence that ranges from 12.9% to 50.9%. If we exclude from our review individual case reports, the migration incidence observed ranges from 1.8% to 57.1% [Table 1]. Studies with longer follow-up have observed a higher incidence of lead migration.^[5] Nonetheless, in this case report, lead migration was detected only 2 months after surgery.

Lead migration is a complication often reported in ONS [Table 1], but most authors have not identified or informed a clear cause for migration. When a presumptive cause was identified, it included a defect in the lead anchor,^[40] iatrogenic lead migration during IPG implantation,^[25] and repeated extension traction due to high mobility in the implanted area.^[26,27,33,36] The association of fibrosis and lead migration has not, to the best of our knowledge, been reported before in literature.

Other factors could also be associated a higher incidence of lead migration. It is reported that cylindrical leads could be more associated with migration than paddle-shaped leads^[10] since the shape and the dimensions of a cylindrical lead make it harder to anchor it to the fascia and increases the rate of skin erosion and lead migration.^[33]

The IPG location can also impact migration. A study described that infraclavicular or low abdomen sites are associated with less pathway length change during flexion and extension than periscapular or gluteal sites and, thus, generate less tension and could be less associated with lead migration.^[38]

Nevertheless, even though we employed techniques to reduce strain on the lead such as forming circular coils with the lead extension, implanting the IPG in an infraclavicular site, not using a cylindrical lead, and anchoring the lead according to manufacturer's specifications, none of these measures could prevent lead migration in the face of fibrosis around the ONS system.

The stress relief loop was positioned in the distal end of the lead extension, in an infraclavicular pocket, alongside the IPG. The positioning of the loop close to the IPG could be

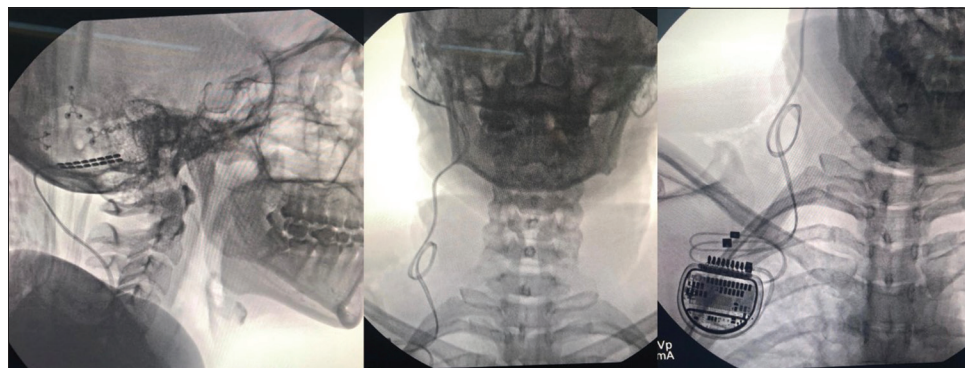


Figure 4: Repositioned lead after surgery.

Table 1: Studies published in the past 10 years reporting lead migration in ONS.

	Study design	Diagnosis	Lead migration incidence (%)	Time until migration
Saper et al. ^[31]	Randomized controlled trial	Chronic migraine	12/51 (23.5)	3 months
Magis et al. ^[21]	Clinical trial	Cluster headache	1/15 (6.6)	7 days
Wolter et al. ^[41]	Clinical trial	Cluster headache	2/7 (28.6)	4.5 months
Abejón et al. ^[11]	Randomized controlled trial	Chronic migraine	4/31 (12.9)	N/A
Vadivelu et al. ^[40]	Case series	Chiari malformation	3/15 (20.0)	N/A
Son et al. ^[36]	Case report	Refractory hypnic headache	1/1 (100.0)	1 year
McGreevy et al. ^[22]	Case report	Occipital neuralgia	1/1 (100.0)	11 months
Serra et al. ^[32]	Prospective, randomized cross-over study	Chronic migraine	3/29 (10.3)	N/A
Mueller et al. ^[25]	Clinical trial	Chronic headache	1/27 (3.7)	46 months
Palmisani et al. ^[27]	Case series	Chronic migraine	2/23 (8.7)	N/A
Silberstein et al. ^[34]	Clinical trial	Chronic migraine	20/157 (12.7)	12 weeks
Lambru et al. ^[17]	Prospective open label	Secondary chronic SUNCT	1/9 (11.1)	N/A
Notaro et al. ^[26]	Case series	Chronic migraine	5/17 (29.4)	2 months
Dodick et al. ^[8]	Randomized controlled trial	Chronic migraine	29/57 (50.9)	N/A
Sharan et al. ^[33]	Randomized controlled trial	Chronic migraine	27/157 (17.2)	96.7 days
Reed et al. ^[30]	Case series	Hemiplegic migraine	2/4 (50.0)	6 months and 22 months
Plazier et al. ^[28]	Case series	N/A	3/92 (3.3)	12 months
Clark et al. ^[7]	Case series	Chronic migraine	7/16 (43.7)	N/A
Magis et al. ^[20]	Case series	Chronic migraine	1/15 (6.6)	N/A
Leone et al. ^[18]	Clinical trial	Cluster headache	6/35 (17.1)	6.5 days
Keifer et al. ^[15]	Case series	Occipital neuralgia	1/29 (3.4)	N/A
Fontaine et al. ^[11]	Observational study	Cluster headache	2/58 (3.4)	N/A
Mekhail et al. ^[23]	Randomized controlled trial	Chronic migraine	3/20 (15.0)	N/A
Miller et al. ^[24]	Cohort	Cluster headache	1/51 (2.0)	N/A
Jones et al. ^[14]	Retrospective review	Chronic daily headaches and occipital neuralgia	12/21 (57.1)	N/A
Aibar-Durán et al. ^[2]	Prospective analysis	Cluster headache	1/17 (5.9)	N/A
Ashkan et al. ^[3]	Case series	Chronic migraine	2/112 (1.8)	N/A
Raoul et al. ^[29]	Case series	Occipital headaches	6/60 (10.0)	N/A
Lepus et al. ^[19]	Prospective registry	Cluster headache	16/105 (15.2)	N/A

ONS: Occipital nerve stimulation, N/A: Not available, SUNCT: Short-lasting unilateral neuralgiform headache with conjunctival injection and tearing

associated with migration, although this relation has not been proposed by other studies before. Some authors suggest that the creation of an additional stress relief loop, producing one in the proximal end and another in the distal end of the extension, could reduce migration.^[10,33]

Another limitation that increases the incidence of migration is that leads produced specifically for ONS are not widely available. Leads designed for spinal cord stimulation are often the only alternative, as were the case with this patient and an issue also reported by other authors.^[10]

Perhaps, an effective alternative to reduce lead migration associated with fibrosis would be a wireless ONS system that does not require an extension. Some studies report the use of such devices,^[4,37,39] but they are not widely available and its usage would require more robust trials.

CONCLUSION

In this case report, the development of fibrosis throughout the ONS system caused constriction points in the stress relief loop that was responsible for lead migration. There is not, to the best of our knowledge, another published description of the association of fibrosis and lead migration.

Lead migration can hinder an effective treatment of occipital neuralgia and demands prompt intervention with lead repositioning. It is important to understand the mechanism responsible for lead migration to prevent it and to reposition the lead in an appropriate manner if it happens.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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