ISSN 1941-5923 © Am J Case Rep, 2017; 18: 1225-1228 DOI: 10.12659/AJCR.905676



Received: 2017.06.07 Accepted: 2017.06.28 Published: 2017.11.20

Authors' Contribution:

Statistical Analysis C

Data Interpretation D Manuscript Preparation E Literature Search F Funds Collection G

Study Design A

Data Collection B

A Case Report of Refractory Notalgia Paresthetica Treated with Lidocaine Infusions

BEF 1 Yaroslava Chtompel ABE 2 Marzieh Eghtesadi ADE 1 Grisell Vargas-Schaffer

1 Department of Anesthesiology and Chronic Pain Management, University Hospital of Montreal (CHUM), Montreal, QC, Canada

2 Department of Chronic Pain and Headache Medicine, University Hospital of Montreal (CHUM), Montreal, QC, Canada

Corresponding Author: Conflict of interest:	Yaroslava Chtompel, e-mail: yaroslava.chtompel@mail.mcgill.ca None declared					
Patient:	Female, 50					
Final Diagnosis:	Notalgia parethetica					
Symptoms:	Hyperalgesia • Pruritus					
Medication:	-					
Clinical Procedure:	Intravenous lidocaine infusion					
Specialty:	Anesthesiology					
Objective:	Unusual or unexpected effect of treatment					
Background:	Notalgia paresthetica is a neuropathic condition that manifests as a chronic itch in the thoraco-dorsal region.					
	It is often resistant to treatment, and specific guidelines for its management are lacking. As such, we present					
	a treatment approach with intravenous lidocaine infusions.					
Case Report:	The case involves a 50-year-old woman with spinal cord injury caused by an epidural abscess. The patient de-					
	veloped notalgia paresthetica and sublesional neuropathic pain following its drainage. In the course of her					
	pain management, she was treated with intravenous lidocaine which resulted in profound relief of notalgia					
	paresthetica.					
Conclusions:	Intravenous lidocaine was effective in relieving neuropathic itch in the patient case presented.					
MeSH Keywords:	Lidocaine • Nerve Compression Syndromes • Neuralgia • Pruritus					
Full-text PDF:	https://www.amjcaserep.com/abstract/index/idArt/905676					



Background

Notalgia paresthetica (NP) is a term that describes a chronic neuropathic itch in the posterior thoracic region. It is unilateral, frequently appears at shoulder blade level and is thought to be secondary to damage of the posterior primary rami of the thoracic nerves T2–T6 [1]. One hypothesis explaining this condition is that the cutaneous branches of these spinal nerves coursing perpendicularly through the multifidus muscle cause them to be impinged, thereby leading to a peripheral sensory neuropathy which can manifest as a persistent itching sensation [2]. However, degenerative changes, which can be seen on imaging of the spine, have also been linked to this syndrome and represent another source of entrapment [3]. The exact incidence of NP is unknown as it remains largely underreported.

The presence of intense itch can lead to scratching and hyperkeratosis of the skin which can be seen on physical examination, while skin biopsy can reveal nonspecific post-inflammatory hyperpigmentation [4]. Other features associated with a neuropathic process can be present as well, such as paresthesia, hypoesthesia, allodynia, or hyperalgesia [1].

Treatments previously described for notalgia paresthetica include topical agents such as lidocaine [5] and capsaicin [6]. Unfortunately, their effect is temporary and requires multiple applications per day which can be difficult for the patient to reach when targeting the interscapular area. Systemic agents used in neuropathic pain conditions such as gabapentin and oxcarbazepine have been describes as well [7–9] but good quality randomized controlled trials are lacking. Onabotulinum toxin A intramuscular injections in the pruritic area were shown to be ineffective [10]. Non-invasive modalities including transcutaneous electrical nerve stimulation (TENS), physical therapy, and osteopathic manipulation have been reported in a small number of patients with inconsistent relief [11–13].

In this article, we present a case of NP that was successfully treated with intravenous lidocaine infusions, a modality frequently used in neuropathic pain conditions but not previously described for pruritic nerve entrapment syndromes.

Case Report

A 50-year-old Caucasian woman was referred to our pain management clinic with an eight year history of back and lower limb pain following a T3–T7 laminectomy. The surgical intervention was performed for drainage of a thoracic epidural abscess in the context of intravenous drug use. The patient suffered partial spinal cord damage caused by the abscess and continued to have spasticity and diffuse neuropathic pain below the lesion. Following rehabilitation, the patient regained full segmental strength in her lower limbs and was ambulating with Canadian crutches. The patient's other comorbidities included longstanding insomnia, generalized anxiety, and gastroesophageal reflux disease. At the age of 37, she underwent anterior C5–C7 fusion for a herniated disc and spinal stenosis, without any neurological deficit following surgery.

Within the first year after abscess drainage, she developed a pruritic patch of skin medial to her right shoulder blade. The itch was very bothersome and the patient underwent an evaluation by a dermatologist. A diagnosis of NP was made without the need for a skin biopsy. A trial of oral antihistamines and topical hydrocortisone cream was unsuccessful. The symptoms of intense itching with compulsive thoughts of scratching her back with a sharp object continued to persist.

Prior to her referral to our center, the patient had undergone successful treatment for her addiction at a detoxification center and was started on maintenance methadone for both addiction and pain.

Over the course of three years since her initial evaluation at our clinic, the management focused on treating her neuropathic pain and spasticity. She was started on pharmacological therapy, including pregabalin 300 mg (orally two times a day), methadone was maintained at 30 mg (orally, three times per day), and nabilone 0.5 mg (orally at bedtime). Her breakthrough medication was acetaminophen 300 mg/codeine 30 mg tablets of which she would take 2–4 tablets per day. To treat her insomnia, she was started on mirtazapine 15 mg orally at bedtime and quetiapine 12.5–25 mg orally at bedtime. For the management of her spasticity, she was taking orphenadrine 200 mg orally at bedtime after failing to respond to cyclobenzaprine, methocarbamol, and baclofen. None of these treatments had any effect on her itch, which she continued to describe as pervasive because it was constant and unreachable.

At our Pain Management Center, a series of intravenous lidocaine infusions were proposed as a treatment plan for her neuropathic sublesional pain syndrome. At that time, she presented three different sources of discomfort. The first was a burning pain in her feet, posterior thighs, and buttocks which she rated as 8/10 on a 0 to 10 numerical pain rating scale (NRS) [14]. She also described stabbing and burning pain in the midline thoracic area as 6/10 on the NRS. The second source of pain corresponded to the surgical incision site. Both sources of pain were independent of spinal mobility and were worsened by light touch or pressure. The third source of discomfort was the itch due to NP which was rated as 3/10 on the NRS, constant, and without any day to night variability. On physical examination of the patient's back, a diffuse allodynia and pressure hyperalgesia was noted over her entire back region. The pruritic patch of skin did not show any hyperpigmentation but



Figure 1. Area of notalgia paresthetica prior to lidocaine infusions. The dashed line represents the area of itch with dimensions of 18 cm by 16 cm (measurement not shown).

several auto-induced scratch marks were visible medial to the tip of the right scapula. The area of pruritus (Figure 1) did not demonstrate hypoesthesia.

She underwent a series of three infusions of lidocaine at two week intervals, at a dose of 1 mg/kg bolus followed by 4 mg/kg infusion over one hour (75 mg bolus and 300 mg infusion for 78 kg female). Following the first two infusions, no change was noted in the level of neuropathic pain. However, a significant improvement of the itch was noted. Specifically, after a 12 hour delay, a 90% improvement was reported lasting seven days after the first treatment and 10 days after the second. When the itch progressively reoccurred, it was less intense than at baseline and reached only 80% of its original severity. After the third infusion, the patient experienced a profound decrease in lower limb pain with a NRS of 1/10 for the duration of 14 days. The thoracic pain remained unchanged. The itch disappeared completely for the duration of three weeks with a progressive return to baseline within the month (Table 1). The patient reported a high level of satisfaction during periods of relief as she had been able to forget about the itch and had good pain control. The patient also noted improved activities of daily living and social activities. However, the dimensions of the pruritic region on examination remained unchanged upon reassessment. Subsequently, lidocaine infusions were continued at four week intervals. She did not experience any adverse effects during the infusions.

Discussion

The pathophysiology of NP, which by definition is a sensory peripheral neuropathy, remains hypothetical at the present time as cases have been reported with and without thoracic nerve entrapment in corresponding dermatomes [15,16]. The patient described in our case exhibited evidence of mild spinal cord atrophy on magnetic resonance imaging at the thoracic level corresponding to the notalgia location. A spinal cord injury in association with neuropathic itch has been described previously [17] but attributing it as a cause of NP in our case remains speculative. Spinal cord atrophy does not explain why the pruritic patch is unilateral despite causing bilateral sublesional neuropathic pain. Possibly, the development of notalgia in our patient could have been secondary to paraspinal muscle retraction intraoperatively or due to its spasm postoperatively which could in turn cause cutaneous nerve branch injury. The pruritic area in our patient extended over several dermatomes, as is usually the case, with involvement ranging from one to six dermatomes [15].

Treatment approaches for NP have been largely adapted from those used for neuropathic pain conditions. Intravenous lidocaine has been extensively utilized for pain management in both acute and chronic settings [18,19]. Systemic lidocaine has been shown to inhibit peripheral and central pain transmission through the blockage of voltage-sensitive sodium channels [20]. However, recent *in vitro* studies demonstrated that calcium and potassium channels, N-methyl-D-aspartate receptors as well as modulation of the immune response, may also be involved [21]. Several meta-analyses demonstrated that systemic lidocaine,

Table 1. Pain levels pre- and post-lidocaine infusions according to pain source.

Source of pain	Pre-lidocaine pain level (NRS)*	Post-1 st lidocaine treatment pain level	Post-2 nd lidocaine treatment pain level	Post-3 rd lidocaine treatment pain level	Maximum duration of relief
Midline thoracic region	6/10	6/10	6/10	6/10	N/A**
Lower extremities	8/10	8/10	8/10	1/10	2 weeks
Notalgia paresthetica	3/10	0/10	0/10	0/10	3 weeks

* NRS, numerical rating scale for pain; ** N/A, not applicable.

both in parenteral and oral formulations, is superior to placebo in treating chronic neuropathic pain [23,23]. The majority of randomized controlled studies looking at neuropathic pain due to various underlying causes have shown 5 mg/kg of intravenous lidocaine to be effective [19]. Specifically in spinal cord injury, Attal et al. achieved a reduction in spontaneous pain and allodynia [24]. The difference in our case was the administration of a bolus dose of 1 mg/kg to achieve a rapid plasma concentration followed by an infusion of 4 mg/kg. The optimal duration of infusion to deliver this dose has not been clearly established and ranged between 30 minutes and six hours in previous studies [19]. The decision to deliver the infusion over 60 minutes was made on the basis of maintaining an adequate plasma concentration while minimizing adverse events which can occur with a rapid lidocaine administration.

The use of intravenous lidocaine in pruritus has been limited to patients with renal failure undergoing hemodialysis and those with cholestatic liver disease [25,26]. To our knowledge, no reports have been previously published that describe using intravenous lidocaine for the management of NP; we performed a search in the PubMed database using keywords: notalgia paresthetica, pruritus, intravenous lidocaine. The hypothesis concerning its efficacy relies on the fact that itch can be the sole manifestation in a neuropathic state, which is what NP is believed to

References:

- 1. Massey EW, Pleet AB: Localized pruritus-notalgia paresthetica. Arch Dermatol, 1979; 115: 982–83
- 2. Massey EW, Pleet AB: Electromyographic evaluation of notalgia paresthetica. Neurology, 1981; 31: 642
- Savk E, Savk SO, Bolukbasi O et al: Notalgia paresthetica: A study on pathogenesis. Int J Dermatol, 2000; 39: 754–59
- 4. Savk O, Savk E: Investigation of spinal pathology in notalgia paresthetica. J Am Acad Dermatol, 2005; 52: 1085–87
- Layton AM, Cotterill JA: Notalgia paraesthetica report of three cases and their treatment. Clin Exp Dermatol, 1991; 16: 197–98
- Wallengren J, Klinker M: Successful treatment of notalgia paresthetica with topical capsaicin: Vehicle-controlled, double-blind, crossover study. J Am Acad Dermatol, 1995; 32: 287–89
- 7. Shin J, Kim YC: Neuropathic itch of the back: A case of notalgia paresthetica. Ann Dermatol, 2014; 26: 392–94
- 8. Wolf Maciel A, Ortiz Laraia I, Rowilson Cunha P, Trevisan F: Efficacy of gabapentin in the improvement of pruritus and quality of life in patients with notalgia paresthetica. An Bras Dermatol, 2014; 89: 570–75
- Savk E, Bolukbasi O, Akyol A, Karaman G: Open pilot study on oxcarbazepine for the treatment of notalgia pareasthetica. J Am Acad Dermatol, 2001; 45: 630–32
- Maari C, Marchessault P, Bissonnette R: Treatment of notalgia paresthetica with botulinum toxin A: A double-blind randomized controlled trial. J Am Acad Dermatol, 2014; 70: 1139–41
- Savk E, Savk O, Sendur F: Transcutaneous electrical nerve stimulation offers partial relief in notalgia paresthetica patients with a relevant spinal pathology. J Dermatol, 2007; 34: 315–19
- 12. Fleischer A, Meade T, Fleischer A: Notalgia paresthetica: Successful treatment with exercises. Acta Derm Venereol, 2011; 91: 356–57
- Richardson B, Way B, Speece A: Osteopathic manipulative treatment in the management of notalgia paresthetica. J Am Osteopath Assoc, 2009; 109: 605–8

be. Therefore, intravenous lidocaine treatments utilized in neuropathic pain may be of benefit. In the case presented, an important decrease in the intensity of itch up to 90% in our patient was attributed to the property of lidocaine to decrease the afferent transmission of the pain as well as the itch impulse. The effect of the treatment was not sustained, however, and the intensity of the itch progressively returned to baseline after a period of three weeks. The implication of this effect could represent the need for maintenance infusions at three to four week periods or a transition to an oral form of lidocaine, in our patient.

Conclusions

In summary, we have reported a case of NP associated with spinal cord injury that responded to intravenous lidocaine treatment. The relief was temporary but significant for a three week period, with improved daily function. The exact pathophysiology of NP remains to be elucidated as well as the role of systemic lidocaine in the treatment of neuropathic itch.

Conflicts of interest

None.

- Jensen MP, Turner JA, Romano JM, Fisher LD: Comparative reliability and validity of chronic pain intensity measures. Pain, 1999; 83: 157–62
- 15. Savk O, Savk E: Investigation of spinal pathology in notalgia paresthetica. J Am Acad Dermatol, 2005; 52: 1085–87
- 16. Eisenberg E, Barmeir E, Bergman R: Notalgia paresthetica associated with nerve root impingement. J Am Aca Dermatol, 1997; 37: 998–1000
- 17. Crane DA, Jaffee KM, Kundu A: Intractable pruritus after traumatic spinal cord injury. J Spinal Cord Med, 2009; 32: 436–39
- Kranke P, Jokinen J, Pace NL et al: Continuous intravenous perioperative lidocaine infusion for postoperative pain and recovery. Cochrane Database Syst Rev, 2015; (7): CD009642
- 19. Kosharskyy B, Almonte W, Shaparin N et al.: Intravenous infusions in chronic pain management. Pain Physician, 2013; 16: 231–49
- Woolf CJ, Wiesenfeld-Hallin Z: The systemic administration of local anaesthetics produces a selective depression of C-afferent fibre evoked activity in the spinal cord. Pain, 1985; 23: 361–74
- 21. Van der Wal SEI, Van der Heuvel SAS, Radema SA et al: The *in vitro* mechanisms and *in vivo* efficacy of intravenous lidocaine on the neuroinflammatory response in acute and chronic pain. Eur J Pain, 2016; 20: 655–74
- 22. Tremont-Lukats IW, Challapalli V, McNicol ED et al: Systemic administration of local anesthetics to relieve neuropathic pain: A systematic review and meta-analysis. Anesth Analg, 2005; 101: 1738–49
- Teasell RW, Mehta S, Aubut JAL et al: A systematic review of pharmacological treatments of pain following spinal cord injury. Arch Phys Med Rehabil, 2010; 91: 816–31
- Attal N, Gaude V, Brasseur L et al: Intravenous lidocaine in central pain: A double-blind, placebo-controlled, psychophysical study. Neurology, 2000; 54: 564–74
- 25. Tapia L, Cheigh JS, David DS et al: Pruritus in dialysis patients treated with parenteral lidocaine. N Engl J Med, 1977; 296: 261–62
- Villamil AG, Bandi JC, Galdame OA et al: Efficacy of lidocaine in the treatment of pruritus in patients with chronic cholestatic liver diseases. Am J Med, 2005; 118: 1160–63