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Treatment of dopplerable nummular headache with minimally invasive arterectomy under local anesthesia

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Summary *Objective:* The objective of the current study is to elucidate the potential role of surgery in the treatment of nummular headache (NH).
Background: NH is a disorder in which pain is localized to a specific area. Treatment has traditionally been medical, with the recent addition of nerve blocks and botox injection with equivocal results.
Design: Forty-nine patients were identified using the International Classification of Headache Disorders, third edition, beta version. Patients were asked to identify the area of maximal pain. Patients who had an associated Doppler signal within the area of pain underwent surgical arterectomy using local anesthesia. Preoperative and postoperative headache frequency, severity, duration, and headache-free days were analyzed.
Results: There were a total of 49 patients included in the study (42F:7M) with an average age of 45 years (21–65 years). The average follow-up period was 16 months with a range of 8–33 months. There was a significant reduction in the frequency (-10.7 days; $p < 0.001$), severity (-3.5; $p < 0.001$), and duration (-0.3 hours; $p = 0.4$) of the headache. There was a significant increase in the number of headache-free days per month (10 vs. 21; $p < 0.001$). Headache index decreased by 39.6%, from an average of 378.6 to 228.4 ($p < 0.05$). Twelve patients (24.5%) were free from NH and able to discontinue their medications. There were no complications identified during the follow-up period.

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Conclusion: NH, although rare, can be associated with significant disability despite current treatment modalities. In select patients, surgical arterectomy is a safe, minimally invasive, and effective treatment for NH.

Introduction

Nummular Headache (NH) is a rare disorder occurring with an estimated incidence rate of 6.4–9/100,000 in a hospital-based series.[1,2] The International Classification of Headache Disorders (ICHD-3), third edition, beta, describes NH as a sharply contoured pain, often chronic, of highly variable duration, in a small, circumscribed area (1 to 6 cm) of the scalp. The area of pain has often a round or elliptical shape, and typically occurs in the parietal region and tends to be "side-locked".[3] Over-the-counter (OTC) analgesics are usually adequate to treat the majority of patients with NH,[2] but there is a subset of patients whose symptoms are recalcitrant, thus necessitating additional modalities and in some cases, continuous prophylaxis.[4] These additional, previously described therapies include tricyclic antidepressants, gabapentin, nerve blocks, and onobotulinum toxin A (Botox®), all with varying efficacies.[5–13] Although there remains controversy regarding a "central" vs. "peripheral" etiology of NH, it is largely thought to be related to a peripheral source. There are a number of case reports and series demonstrating patients who have developed or had their NH exacerbated by associated vascular lesions.[14]

The lead author (B.G.) has extensive experience in the surgical treatment of migraine headaches (MH). In particular, peripheral trigger sites have been successfully treated using surgical arterectomy with local anesthesia.[15] This has been further improved by including a preoperative Doppler examination to identify the surgical target.[16] Therefore, it was postulated whether surgical treatment could also be effective in the treatment of NH. Thus, the objective of the current study was to assess whether surgical arterectomy under local anesthesia could be effective in the treatment of NH.

Methods

Forty-nine patients were identified using the International Classification of Headache Disorders classification in the lead author's practice by a trained, board-certified neurologist (Co-author D.R.). Site-specific preoperative NH frequency, severity, duration, and headache-free days were obtained. Before undergoing surgery, patients were asked to identify the area of maximum pain. All patients had an associated Doppler signal within the area of pain and subsequently underwent surgical arterectomy by administering local anesthesia. For some patients, the ultrasound Doppler was maneuvered around within several millimeters of the area of pain indication to detect the Doppler signal. In most patients, the Doppler signal was identified with ease and at the exact site where the patients identified pain with an index finger. In patients who had headache at the time of examination, a nerve block was achieved with almost invariable success. However, a negative result of nerve block did not deter the lead author from proceeding with surgery.

After the surgical site was identified and consent obtained, the patient was brought to the procedure room and placed on an adjustable procedure chair. Patients with anxiety were administered preoperatively with either a narcotic or anxiolytic agent. However, the overwhelming majority of the patients did not require any additional treatments. The patient's face was prepped and draped using a dilute solution of Betadine and sterile drapes/towels. For those with pain in the hair-bearing region, a small area around the Doppler signal site was shaved using surgical clippers. The previously marked site of pain and associated Doppler signal was injected with lidocaine 1% with epinephrine (1:100,000). For patient comfort, a small, half-inch, 30-gauge needle was used and the injection was administered slowly with single skin penetration. The patients who had pain at the time of surgery were asked to report their pain level both before and after injection on a scale of 1–10. However, not necessarily, pain relief following injection was considered a positive prognostic factor. While awaiting medication onset, an incision of approximately 0.6 to 1.2 cm in length was made over the identified Doppler signal area. A No. 15 blade was used to make an incision through the epidermis and dermis of the skin. Dissection was performed using a 0.5-cm mosquito hemostat. Once the artery was identified, it was dissected both proximally and distally to obtain a segment for removal. Larger arteries required ligation using 5-0 or 6-0 Monocryl sutures, whereas smaller arteries were adequately treated by targeted cauterization. The entire intervening segment of the artery and small, intimately associated nerves were removed. Larger nerves required to be divided and the cut end was buried into the temporalis muscle to minimize the potential for a neuroma formation. Following meticulous hemostasis, the incision was closed, and typically, a space obliterating stitch was placed first. In the hair-bearing areas, a 5-0 plain gut suture was used, whereas in more cosmetically sensitive areas, a 6-0 fast gut was used (**Video 1**). Antibiotic ointment was applied on the incision and thereafter, patients were advised to apply two times a day for 5 to 7 days. Patients were allowed to take shower and cleanse the area gently after 24–48 hours. Patients were allowed to return to work and resume routine activity the same day or the day after the procedure. Narcotic medication was rarely necessary. Infrequently, patients experienced an acute worsening in their pain the night following the procedure, but this was rare and self-limiting. Patients refrained from nonsteroidal anti-inflammatory drug (NSAID) use one week before and two to three weeks after the procedure. Patients were typically followed up for one day, one week, one month, three months, six months, and one year postoperatively. Postoperative follow-up communication was set up through FaceTime, Skype or phone for out-of-town patients. Data of postoperative headache frequency, severity, duration, and

headache-free days were obtained. Headache index was calculated by multiplying frequency by severity and length of headache. The number of headache-free days was also assessed. Patient charts were reviewed for complications. Statistical analysis was performed using Student's t-tests.

Results

There were a total of 49 patients included in the study (42F:7M) with an average age of 45 years (21–65 years). The average follow-up was 16 months with a range of 8–33 months (Table 1). Patients presented with predominant headache on the right side for 55% of the time and on the left for 44% of the time. In total, 65.3% of patients underwent bilateral procedures as well as 22.4% and 12.2% underwent right-sided and left-sided auriculotemporal decompression procedures, respectively, for treatment of NH; 7.2% of patients underwent NH surgical treatment alone, whereas the remaining 92.8% of patients also underwent concomitant or previous migraine surgeries. NH frequency significantly decreased by an average of 10.7 days, from 19.7 to 9 days (54.3% reduction; $p < 0.001$) (Figure 1). There was a significant increase in the number of headache-free days per month (10 vs. 21; $p < 0.001$) (Figure 2). NH severity was significantly improved with an average reduction of 3.5 from 8.2/10 to 4.7/10 based on the visual analog scale ($p < 0.001$) (Figure 3). NH duration decreased by 0.3 hours ($p = .4$) (Figure 4). Headache index decreased by 39.6%, from 378.6 to 228.4 ($p < .05$) (Figure 5). Twelve (24.5%) patients were free from NH and able to discontinue their medications. An additional 22 patients (45%) reported at least a 50% reduction in their headache index scores. This means that in addition to patients who were free from headache, approximately 70% of the patients in this series had at least a 50% reduction in their headache severity scores (Figure 6). There were no complications identified during the follow-up period.

Table 1 Patient demographic and follow-up data.

Sex	
Male	7
Female	42
Mean Age	45 years (range 21–65)
Laterality	
Right	55%
Left	45%
Mean Follow-up	16 months (range 8–33)

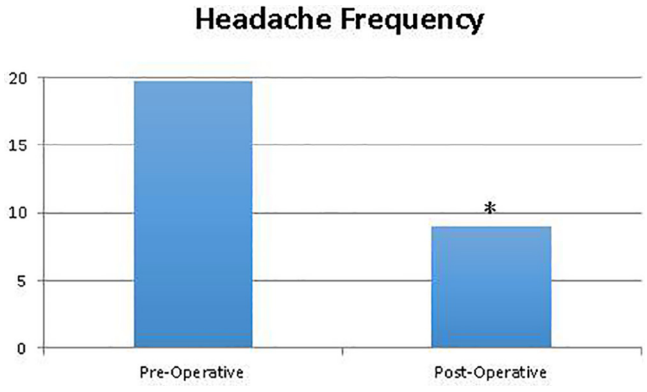

Figure 1 Preoperative and postoperative NH frequency. NH frequency was significantly reduced by an average of 10.7 days from 19.7 to 9 days (54.3% reduction; $p < 0.001$). * indicative of statistical significance.

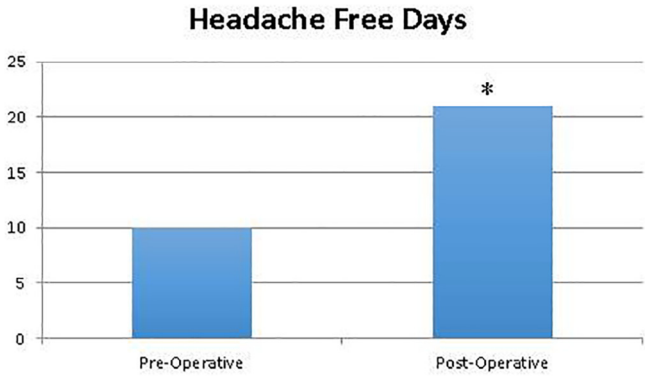

Figure 2 Preoperative and postoperative headache-free days. There was a significant increase in the number of headache-free days per month (10 vs. 21) * indicative of statistical significance.

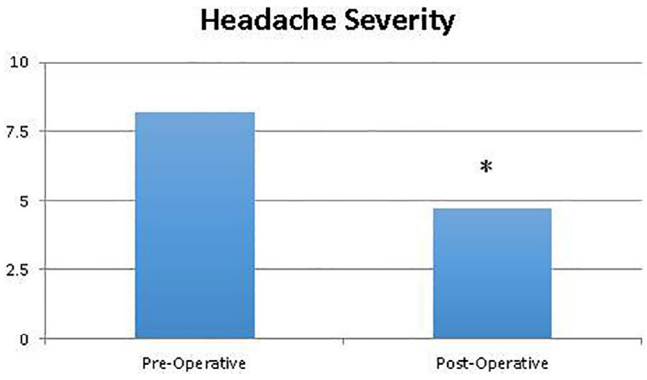

Figure 3 Preoperative and postoperative NH severity. NH severity was significantly improved with an average reduction of 3.5 from 8.2/10 to 4.7/10 based on the visual analog scale ($p < 0.001$). * indicative of statistical significance.

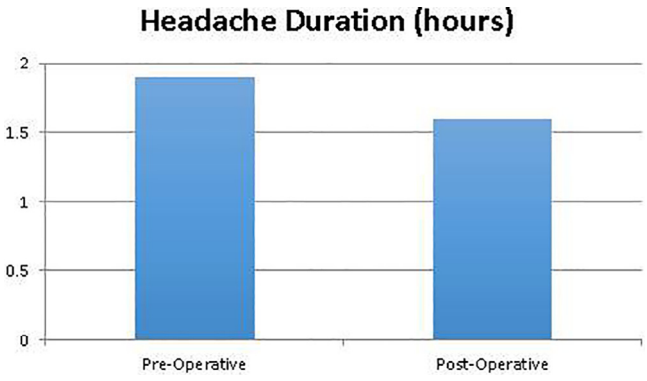

Figure 4 Preoperative and postoperative NH duration. NH duration decreased by 0.3 hours ($p = .4$).

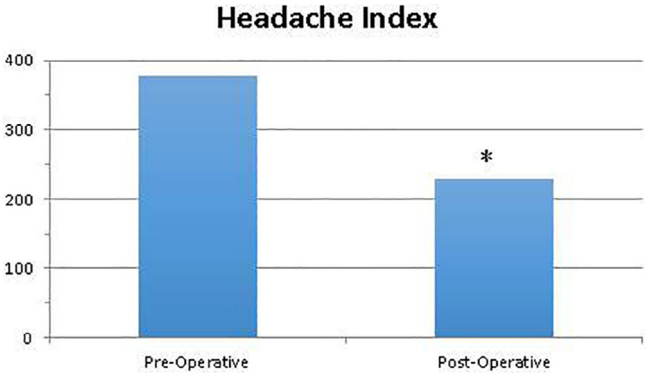

Figure 5 Preoperative and postoperative headache index. Headache index was calculated by multiplying frequency by severity and length of headache. Headache index decreased by 39.6%, from 378.6 to 228.4 ($p < .05$). * indicative of statistical significance.

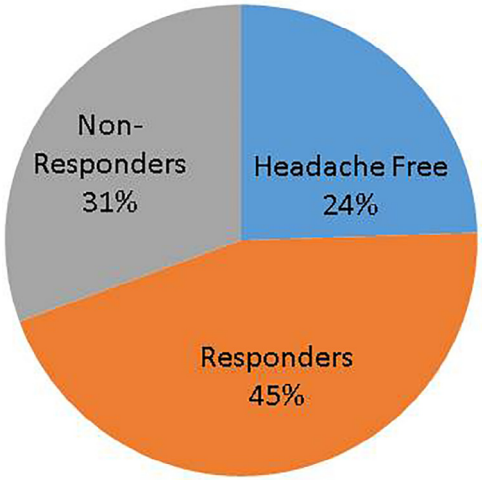

Figure 6 Patient disease status after one surgical intervention for nummular headache at time of the latest follow-up. Fifteen patients (30.6%) were classified as nonresponders; 22 (44.9%) were classified as responders, defined as a reduction in headache index score by 50%; and 12 (24.5%) were classified as those free from headache, not requiring any medication for headache.

Discussion

Arterectomy for the elimination of vascular irritation of the nerves that are responsible for painful conditions such as trigeminal neuralgia and MH has been previously reported.[17] This is the first study of its kind to address the potential surgical treatment of NH. NH, although rare, can have potentially debilitating effects on affected individuals. Until now, treatment strategies were mainly focused on medications with inconsistent efficacies. The use of gabapentin for NH demonstrated only a partial benefit in 60% of patients.[3] Tricyclic antidepressants such as amitriptyline, nortriptyline, and protriptyline have also been tried in more than 40 published cases, and these also provided only a partial benefit in 45% of patients with NH.[3] Interestingly, peripheral nerve blocks have shown disappointing results, thus demonstrating minimal to no relief in 42% of a 40-patient case-report,[3] whereas the use of botulinum toxin type A has demonstrated some efficacy in a case series of 24 patients, with 92% achieving at least some benefit.[5,6,8,18] There have been additional case reports on the use of indomethacin, carbamazepine, neurotropin, and cyclobenzaprine, but these are yet to be substantiated.[9–11,13] Treatment with transcutaneous electrical nerve stimulation has also been reported as being effective in one case.[12] Medications can be expensive and are not without associated side effects. All these treatments including botulinum toxin require continued administration to provide long-term benefit.

Our current study demonstrates a significant reduction in NH frequency and severity following minimally invasive surgical arterectomy under local anesthesia. There was a statistically significant increase in the number of headache-free days from 10 to 21 days. Moreover, nearly one-fourth (24.5%) of the patients were free from headache after one operation and were not requiring medication for headache during the completion of this study. NH frequency and severity were reduced by 54.3% and 42.6%, respectively. An additional 22 patients (45%) reported at least a 50% reduction in their headache index scores. This result shows that in addition to the patients who were free from headache, approximately 70% of the patients in this series had at least a 50% reduction in their headache severity score. Although there were 5 (10%) patients who reported increased frequency and decreased headache-free days following surgery, we have observed that the overwhelming majority of the residual headaches are almost never exactly on the same site and are often in a different, remote area or at least several millimeters away from the initial incisional scar. The latter condition may denote redirection of the blood flow in another branch of the blood vessel, thus resulting in nerve irritation. These results, however, demonstrate superior treatment outcomes when compared to the currently available options and are in stark contrast to the aforementioned case reports of little to minimal benefit for a wide range of medications. Although there are risks associated with surgery, this surgical procedure is minimally invasive and does not require general anesthesia. Most patients either take only one or two analgesics on the night of surgery or do not require any medications. The recovery time is minimal and patients can return to work the same day or the next day. Positive results are commonly experienced immediately after surgery. Patients may have some postoperative surgical discomfort, rather than symptoms associated with NH, and they are able to identify and describe the difference. In the current cohort, there were no complications in the interim follow-up period.

Interestingly, surgical arterectomy for the treatment of NH may have some, yet to be completely elucidated, physiological basis. Similarly, the role of a vascular nidus for migraines, namely, the trigeminovascular system, has been postulated, but not yet entirely explored.[19] There are several reports of *de novo* onset of NH in patients developing pain in close proximity to sites of aneurysms or calcified hematomas.[14,20] There is also some indication that triptans, which cause vasoconstriction, are somewhat effective.[14] These associations suggest a vascular component to NH, which is further supported by the current study. In a prior study, patients with MH demonstrated a relative lack of myelin by electron microscopy compared to patients without migraines. It could be postulated that the relative lack of myelin, and

therefore insulation, could make the nerves more sensitive to perivascular irritation, thereby resulting in a localized neuritis and associated headaches. This may also explain the relatively high incidence of MH in our patient population, as MH and NH could have a common etiology.[21] A recently accepted and soon-to-be-published study also supports that an interaction between the artery and nerve is critical to the etiology and surgical treatment of headaches. Although none of the patients in this group are known to have undergone a revision, we have recently demonstrated in a randomized controlled trial that migraine patients who did not undergo an arterectomy had a revision rate of 31%. This rate was found to be significantly higher than the overall revision rate of 8.7% obtained for the same surgery by the senior author in 978 patients over a 15-year period. Additional studies investigating differences between arteries surgically removed from patients with and without headaches, as well as inflammatory markers, would be an interesting topic of discussion. However, it is currently beyond the scope of this submission.

The current study is not without limitations. The sample size may be underpowered to eliminate other potential factors. However, this is one of the larger cohorts to be presented in literature. Additionally, because of the rare nature of the disease, it would be extremely difficult and potentially impossible to recruit adequate patients to eliminate all potential confounding variables. Another potential limitation is the possibility of a "placebo effect". Although we cannot entirely eliminate this, the exceptional response rate and long-term effect would contradict such an effect. Theoretically, a sham surgery study could be designed, in which patients may be randomly selected to undergo either arterectomy or sham surgery without arterectomy. Patients would be blinded to the procedure that would be performed. In addition to the numerous ethical dilemmas this study poses, logically, given the overall incidence of NH, this is not technically feasible. Finally, although these patients were identified using standardized criteria by a trained neurologist, there was a higher incidence of heterogeneity with regard to NH, with and without migraine. Recent studies have attempted to develop a differentiation between NH and atypical NH, but there is no current consensus.[22] NHs, both with and without migraines, are most likely to include a spectrum of illnesses that is yet to be completely elucidated.

Conclusions

NH, although rare, can be associated with significant disability despite current treatment modalities. In select patients, surgical arterectomy is a safe, minimally invasive, and effective treatment for NH with superior outcomes compared to other currently available options.

Funding

None.

Conflicts of interest

None declared.

Ethical approval

IRB approval #07-13-23.

References

1. Pareja JA, Pareja J, Barriga FJ, et al. Nummular headache: a prospective series of 14 new cases. *Headache* 2004;**44**:611–4.
2. Grosberg BM, Solomon S, Lipton RB. Nummular headache. *Curr Pain Headache Rep* 2007;**11**:310–2.
3. Schwartz DP, Robbins MS, Grosberg BM. Nummular headache update. *Curr Pain Headache Rep* 2013;**17**:340.
4. Pareja JA, Montojo T, Alvarez M. Nummular headache update. *Curr Neurol Neurosci Rep* 2012;**12**:118–24.
5. Ruscheweyh R, Buchheister A, Gregor N, Jung A, Evers S. Nummular headache: six new cases and lancinating pain attacks as possible manifestation. *Cephalalgia* 2010;**30**:249–53.
6. Dusitanond P, Young W. Botulinum toxin type A's efficacy in nummular headache. *Headache* 2008;**48**:1379.
7. Matthew N, Kailasam J, Meadors L. Botulinum toxin type A for the treatment of nummular headache: four case series. *Headache* 2008;**48**:442–7.
8. Seo M, Park S. Botulinum toxin in nummular headache. *Cephalalgia* 2005;**25**:991.
9. Baldacci F, Nuti A, Lucetti C, Borelli P, Bonuccelli U. Nummular headache dramatically responsive to indomethacin. *Cephalalgia* 2010;**30**:1151–2.
10. Man YH, Yu TM, Li LS, Yao G, Mao XJ, Wu J. A new variant nummular headache: large diameter accompanied with bitrigeminal hyperalgesia and successful treatment with carbamazepine. *Turk Neurosurg* 2012;**22**:506–9.
11. Yamazaki Y, Kobatake K. Successful treatment of nummular headache with Neurotropin(R). *J Headache Pain* 2011;**12**:661–2.
12. Tayeb Z, Hafeez F, Shafiq Q. Successful treatment of nummular headache with TENS. *Cephalalgia* 2008;**28**:897–8.
13. Robbins MS, Grosberg BM. Menstrual-related nummular headache. *Cephalalgia* 2010;**30**:507–8.
14. Lopez-Ruiz P, Cuadrado ML, Aledo-Serrano A, Alonso-Ovies A, Porta-Etessam J, Ganado T. Superficial artery aneurysms underlying nummular headache–2 cases and proposed diagnostic workup. *Headache* 2014;**54**:1217–21.
15. Guyuron B, Tucker T, Davis J. Surgical treatment of migraine headaches. *Plast Reconstr Surg* 2002;**109**:2183–9.
16. Guyuron B, Nahabet E, Khansa I, Reed D, Janis JE. The current means for detection of migraine headache trigger sites. *Plast Reconstr Surg* 2015;**136**:860–7.
17. Murillo CA. Resection of the temporal neurovascular bundle for control of migraine headache. *Headache* 1968;**8**:112–7.
18. Mathew NT, Kailasam J, Meadors L. Botulinum toxin type A for the treatment of nummular headache: four case studies. *Headache* 2008;**48**:442–7.
19. Burstein R, Noseda R, Borsook D. Migraine: multiple processes, complex pathophysiology. *J Neurosci* 2015;**35**:6619–29.
20. Ulivi M, Baldacci F, Vedovello M, et al. Localized calcific hematoma of the scalp presenting as a nummular-like headache: a case report. *Headache* 2014;**54**:370–2.
21. Guyuron B, Yohannes E, Miller R, Chim H, Reed D, Chance MR. Electron microscopic and proteomic comparison of terminal branches of the trigeminal nerve in patients with and without migraine headaches. *Plast Reconstr Surg* 2014;**34**:796e–805e.
22. Barón J, Rodriquez C, Ruiz M, et al. Atypical nummular headache or circumscribed migraine: the utility of pressure algometry. *Pain Res Manag* 2015;**20**:60–2.