

Pearls & Oy-sters: Postpipeline Headache Phenomenon

Nummular Headache Presenting After Intracranial Aneurysm Stenting

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Neurology® 2022;99:718-720. doi:10.1212/WNL.0000000000201157

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Abstract

We report on a 31-year-old right-handed woman with a medical history of presyncopal episodes and migraine headaches who presented to the outpatient clinic with a nummular headache after intracerebral stenting, which was different than her previous migraines. This represents postpipeline embolization headache phenomenon, which is a relatively new term to describe a new or different headache in individuals who recently underwent intracranial vascular stenting as a treatment for cerebral aneurysms.

Pearls

- Nummular headache is a headache disorder characterized by focal head pain in a well-circumscribed oval or elliptical-shaped area.
- Postpipeline embolization headache is a recently described headache phenomenon that should be considered on the differential diagnosis for patients presenting with new-onset headaches after intracranial flow-diversion stenting.

Oy-sters

- While nummular headache is considered a primary headache disorder, a similar headache phenotype can occur secondary to lesions of the bone, scalp, and underlying intracranial structures.
- New headache following flow-diversion stenting could be related to in-stent stenosis or branch occlusions of perforating arteries, so additional evaluation is recommended.

Case Report

A 31-year-old right-handed woman with a medical history significant for presyncopal episodes and migraine headaches presented to the outpatient clinic for a new-onset persistent headache occurring immediately after intracerebral aneurysm stenting.

She described a long history of infrequent migraine headaches that had escalated in frequency and severity, without provocation. An MRI examination of the brain with and without contrast and a subsequent MR angiogram head without contrast revealed an unruptured saccular aneurysm measuring 5 mm (height) × 4 mm (width) × 4 mm (neck), arising from the paraclinoid region of the right internal carotid artery (ICA). Two to 3 months after her headaches had increased in frequency, she had a cerebral angiogram with placement of a flow-diverting stent in the right ICA across the neck of the aneurysm.

Immediately after the procedure, she developed a new type of headache that was entirely different from her previous migrainous headaches. Instead of intermittent pain, she described a persistent sharp pain localized to an oval shape, approximately 1 inch wide and 3 inches long (longer over the anterior-posterior dimension) in the right parietal region. There was no rash in

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the region and no skin/hair change. The pain was present all day every day with associated allodynia, frequent paresthesia, and occasional sharp stabs in the oval area of pain. Approximately 20 days per month, she would have brief episodes lasting seconds, where she would feel the paresthesia spread from the oval area to the hemicranium or the whole head. She would sometimes feel subjective warmth in the skin of the scalp in that region. She had not noticed any associated tenderness over the eyebrows or skull base. In the clinic, her neurologic examination was unremarkable. She underwent several imaging studies, including CT of the head without contrast, CT angiogram of the head and neck with and without contrast, MR angiogram of the head without contrast, and MR venogram, which were all unremarkable for acute pathologies. She also underwent a cerebral angiogram, which showed stabilization of her implanted flow-diverter device.

The patient was seen in our clinic 3 years after the onset of this persistent focal headache. For abortive headache therapies, she had previously tried butalbital approximately 2 days per week, acetaminophen (approximately 4 days per week), indomethacin (75 mg 3 times daily × 3 months), cyclobenzaprine, and ibuprofen without relief. For preventative headache therapies, she previously tried amitriptyline (small dose), topiramate (did not tolerate), and gabapentin (did not tolerate), without significant relief. During the clinic visit, she was diagnosed with nummular headache and started on a retrial of amitriptyline with a plan of alternative preventatives, including onabotulinumtoxinA, if she continued to have daily pain. During a follow-up visit, she noted that the 50 mg of amitriptyline daily was ineffective, but her response was confounded by her recent daily overuse of acetaminophen tablets and twice weekly use of butalbital/acetaminophen/caffeine tablets. Recommendations for the patient included initiation of onabotulinumtoxinA, continuation of 50 mg of amitriptyline daily, and weaning of daily over-the-counter pain medications.

Discussion

The best term to describe the patient's new headache presentation is postpipeline embolization headache phenomenon. This relatively new term describes a new or different headache in individuals who recently underwent intracranial vascular stenting as a treatment for cerebral aneurysms.¹ A recent study performed a telephone survey of 88 individuals who underwent flow-diverting/Pipeline stenting for the treatment of cerebral aneurysms.¹ Of the patients surveyed, 55% reported a new postprocedural headache with varying characteristics and associated symptoms. Individuals described their pain as dull, sharp, or throbbing, typically located ipsilaterally to the aneurysm/stent. The headaches started an average of 20 days after the procedure, and 69% of patients were still experiencing the headache an average of 21.6 months later. Most patients described intermittent rather than constant head pain, with an average headache frequency of 2–3 times per week, an average duration of 9.7 hours and an average 3.6/10 pain intensity.¹ It is of interest that predictors for developing postpipeline

embolization headache include young age and a history of headaches, both of which were present in our patient.

Numerous headache phenotypes can occur after postflow diverter intracranial stenting.¹ By her description, our patient's postflow diverter headache most closely resembled the phenotype of a nummular headache (NH), characterized by focal head pain in a well-circumscribed oval or elliptical-shaped area. The current *International Classification of Headache Disorders, Third edition (ICHD-3)* diagnostic criteria for NH require the pain to be as follows: (1) continuous or intermittent; (2) felt exclusively in the area of the scalp described as sharply contoured, fixed in size and shape, round/elliptical, and 1–6 cm in diameter; and (3) not corresponding better to another ICHD-3 diagnosis.²

The pain of NH is most often persistent, similar to our case, with up to 75% of published cases describing pain present for more than 3 months.² The painful area may be anywhere on the scalp but is typically located in the parietal region.² Pain is generally described as pressure-like, sharp or stabbing, and is of mild-to-moderate intensity, with occasional superimposed severe exacerbations.³ In a review of 250 patients with NH, the average age at onset was 45 years, with a female predominance.⁴ Similar to our patient, patients with NH commonly have abnormal sensation in the affected area and may describe variable combinations of paresthesia, dysesthesia, and allodynia in the affected area.² Treatment of NH resembles treatment for other primary headache disorders, with 55% of patients requiring preventative therapy.⁵ Gabapentin, indomethacin, tricyclic antidepressants, and onabotulinumtoxinA have been reported effective in select cases.^{3–5} Unfortunately, NH can become refractory to abortive and preventive analgesic therapies, as seen in our patient's presentation.

While NH is considered a primary headache disorder, numerous possible secondary etiologies have been described (Table). Headaches resembling NH have occurred after trauma and in association with bony lesions such as fibrous dysplasia, craniosynostosis, Paget disease, and Langerhans histiocytosis.³ Some secondary cases have described pathology corresponding to the affected area, such as varicella zoster viral infection, calcified hematoma, and linear scleroderma; other cases have a less clear association with the location of pathology, such as contralateral NH after a pituitary adenoma resection.³ Pertinent to our case, there have been several cases of NH associated with vascular pathologies, although each of these were associated with fusiform areas of superficial vessels near the area of pain.³

Our patient developed ipsilateral NH after placement of a flow-diverting stent in the right paraclinoid ICA. Pain from stimulation within the ICA is not without precedent. In classic studies performed by Ray and Wolff⁶ in 1940, the intracranial carotid artery was found to be sensitive to pain brought on by stretching, stroking, and faradic stimulation. Focal referred pain has also been demonstrated ipsilateral to balloon inflation within the distal ICA.⁷ While the intracranial portion of

Table Reported Secondary Etiologies of Nummular Headache^{3,9,10}

Scalp
Subcutaneous cyst
Linear scleroderma
Varicella zoster viral infection
Calcified hematoma of scalp
Insect bite in the affected region
Bone
Bone hemangioma
Fibrous dysplasia
Craniosynostosis
Langerhans histiocytosis
Paget disease
Osteoma
Cholesterol cyst in the bone
Intracranial
Arachnoid granulation
Pituitary tumor (improved after resection)
Cranial surgery; trans-sphenoidal resection of pituitary tumor
Arachnoid cyst
Subtentorial meningioma
Intracerebral cavernoma
Vascular
Superficial artery aneurysms
Postvascular stenting for aneurysm (current case)
Other
Head trauma

the ICA is generally believed to refer pain to the fronto-temporal region due to innervation from the trigeminal nerve, there is significant anatomic variation in the innervation of intracranial structures, with overlapping innervation by both trigeminal and the upper cervical nerves.⁸

Endovascular treatment of aneurysms using flow-diverting devices such as Pipeline stents is becoming relatively common. Most complications are asymptomatic; however, severe complications can also occur including in-stent stenosis or potential branch occlusions of perforating arteries with a risk of ischemia. Therefore, any patient presenting with new unexplained headache after an endovascular stent procedure for an intracranial aneurysm requires additional evaluation. If the evaluation shows no structural abnormalities and the new postprocedural headache remains unexplained, postpipeline embolization headache should be considered in the differential.

Acknowledgment

Brandon Ghislain is deceased. This work is dedicated to the memory of Brandon Ghislain.

Study Funding

No targeted funding reported.

Disclosure

The authors report no relevant disclosures. B. Ghislain is deceased—disclosures not included for this author. Go to [Neurology.org/N](https://www.neurology.org/N) for full disclosures.

Publication History

Received by *Neurology* February 23, 2022. Accepted in final form July 11, 2022. Submitted and externally peer reviewed. The handling editor was Roy Strowd III, MD, MEd, MS.

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Brandon Ghislain, BS		Drafting/revision of the manuscript for content, including medical writing for content; major role in the acquisition of data
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Neurology 2022;99;718-720 Published Online before print August 26, 2022

DOI 10.1212/WNL.0000000000201157

This information is current as of August 26, 2022

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