Pears & Oy-sters: Postpipeline Headache Phenomenon

Nummular Headache Presenting After Intracranial Aneurysm Stenting

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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 post-pipeline 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...
acetaminophen/ca
acetaminophen tablets and twice weekly use of butalbital/
her response was confounded by her recent daily overuse of
50 mg of amitriptyline daily, and weaning of daily over-the-
described intermittent rather than constant head pain, with an
the headache an average of 21.6 months later. Most patients
after the procedure, and 69% of patients were still experiencing
pain as dull, sharp, or throbbing, typically located ipsilaterally to
individuals described their pain as dull, sharp, or throbbing, typically located ipsilaterally to
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, in-
cluding 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 un-
remarkable 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), indo-
trimethacin (75 mg 3 times daily × 3 months), cyclobenzaprine,
and ibuprofen without relief. For preventative headache ther-
pies, 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 retial 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,
her response was confounded by her recent daily overdose of
acetaminophen tablets and twice weekly use of butalbital/
acetaminophen/caffeine tablets. Recommendations for the pa-
ient 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 pre-
sentation 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. A
recent study performed a telephone survey of 88 individuals who underwent flow-diverting/Pipeline stenting for the treat-
ment of cerebral aneurysms. Of the patients surveyed, 55% reported a new postprocedural headache with varying charac-
teristics 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. 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. By her description, our pa-
tient’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 Head-
ache 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 de-
scribed 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.

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. The painful area may be anywhere on
the scalp but is typically located in the parietal region. Pain is
generally described as pressure-like, sharp or stabbing, and is
of mild-to-moderate intensity, with occasional superimposed
severe exacerbations. In a review of 250 patients with NH,
the average age at onset was 45 years, with a female pre-
dominance. Similar to our patient, patients with NH com-
monly have abnormal sensation in the affected area and may
describe variable combinations of paresthesia, dysesthesia, and
allodynia in the affected area. Treatment of NH resembles
treatment for other primary headache disorders, with 55% of
patients requiring preventative therapy. Gabapentin, indo-
trimethacin, tricyclic antidepressants, and onabotulinumtoxinA
have been reported effective in select cases. 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, cranio-
synostosis, Pagen disease, and Langerhans histiocytosis. 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. 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.

Our patient developed ipsilateral NH after placement of a
flow-diverting stent in the right paracaninal ICA. Pain from
stimulation within the ICA is not without precedent. In classic
studies performed by Ray and Wolff 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 inf-
lation within the distal ICA. While the intracranial portion of
the ICA is generally believed to refer pain to the frontotemporal 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.\textsuperscript{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.

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**References**

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