Reminder of important clinical lesson

Chronic daily headaches secondary to greater auricular and lesser occipital neuromas following endolymphatic shunt surgery

Leon Vorobeichik,¹ Michael A Fallucco,² Robert R Hagan²¹Saint Louis University School of Medicine, Saint Louis, Missouri, USA²Neuropax Clinic Peripheral Nerve Surgery, Saint Louis, Missouri, USA

Correspondence to Robert R Hagan, info@neuropaxclinic.com

Summary

In head and neck surgery, peripheral sensory nerves are at risk for traumatic injury. These injuries are known to be peripheral triggers of chronic headaches if left untreated or unrecognised. We report the case of a patient that presented with a severe headache, nausea and emesis of 2 years duration following endolymphatic shunt placement for Meniere's disease. Nerve blockade suggested a peripheral trigger, and surgical exploration of the incision site revealed traumatic neuromas of the greater auricular and lesser occipital nerves. Subsequent nerve resection yielded complete symptomatic relief. This is the first case report of a peripherally triggered migraine headache due to the development of neuromas of the greater auricular and lesser occipital nerves, also representing a previously unreported complication of endolymphatic shunt placement. It is recommended that in patients presenting with intractable migraines and a history of head and neck surgery, diagnostic nerve blockade be used to assess for neuromas.

BACKGROUND

Meniere's disease is characterised by vestibulochochlear symptoms which include intermittent episodes of vertigo lasting from minutes to hours, with fluctuating sensorineural hearing loss, tinnitus and aural pressure.¹ The pathophysiological basis for the disease is the development of an endolymphatic hydrops due to either the obstruction of the endolymphatic duct or dysregulation of endolymph production.² In patients with intractable Meniere's, one potential surgical approach is endolymphatic sac surgery that involves the placement of a mastoid shunt to allow for drainage of the endolymph, with the purpose of restoring proper pressure in the sac.

A review of over 2000 endolymphatic sac procedures demonstrated the success of the procedure with low morbidity. Nearly 75% of the patients experienced complete alleviation of vertigo and vestibular symptoms, and an overall improvement in 90%.³ The main complications were a relatively low incidence of hearing loss, as well as intraoperative bleeding and drainage of spinal fluid.

Neurological complications are rare, with only one report of trigeminal neuralgia.⁴ Here, we present the first case report of peripherally triggered migraine headaches due to the development of traumatic neuromas of the greater auricular and lesser occipital nerves following endolymphatic shunt placement.

CASE PRESENTATION

A 47-year-old woman with a history of Meniere's disease treated by bilateral endolymphatic sac surgery presented to our peripheral nerve surgery clinic with severe intractable migraine of 2 years duration and no history of headache prior to her surgery. The pain generator was in the postauricular and anterior occipital region, causing nausea and occasional emesis and vertigo. Prior to our consultation,

the patient underwent numerous emergency department (ED) admissions as well as a formal neurology evaluation, with an ICHD-II diagnosis consistent with migraine without aura plus chronic migraine. Her symptoms were recalcitrant to nearly 2 years of pharmacological therapy, including a regimen consisting of topiramate 150 mg twice a day, excedrin (acetaminophen/aspirin/cafeine) and ibuprofen, as well as to dilauid and zofran administered during ED admissions. Her headaches were causing severe disability as determined by the Migraine Disability Assessment (MIDAS) with a preoperative score of 215, that being in the severe class of disability.

INVESTIGATIONS

Our physical examination revealed exquisite tenderness to palpation along the superior aspect of the sternocleidomastoid muscle. We subsequently performed a site-specific diagnostic local blockade of the bilateral greater auricular and lesser occipital nerves using a mixture of 0.25% marcaine and 1% lidocaine, with 5 cc per injection site. The diagnostic blockade was successful in alleviating the patient's headache pain during the duration of the anaesthetic.

TREATMENT

Subsequently, under general anaesthesia, the patient was positioned in the supine position enabling access to the postauricular region. A 3 cm linear incision was made at the posterior and superior border of the sternocleidomastoid muscle extending into the hairline. Using a nerve stimulator to ensure protection of the spinal accessory nerve, the great auricular and lesser occipital nerves were located. Neurolysis was performed from their origins at the posterior border of the sternocleidomastoid muscle distally into dense scar tissue at the site of previous incision for endolymphatic sac surgery. The quality of the

nerves within the scar tissue did not make neurolysis possible and therefore resection and burial of the proximal nerve segments into the sternocleidomastoid muscle was performed bilaterally.

OUTCOME AND FOLLOW-UP

Postoperatively the patient began range-of-motion neck exercises without difficulty. There were no wound-healing complications. Throughout the 1-year follow-up period, the patient reported complete resolution of her previous chronic daily headache, with a postoperative MIDAS Score of 8 and without the need for the continuation of pharmacological therapy.

DISCUSSION

We present the case report of intractable headache development secondary to peripheral neuroma formation following endolymphatic sac surgery, as well as its successful treatment. While migraine headaches are traditionally thought to be centrally mediated, they may also emanate from peripheral trigger sites,⁵ with peripheral nerve blockade emerging as successful therapy.⁶ As illustrated by this report, neuromas may provide the impetus for such a migraine trigger. Traumatic neuromas represent reactive hyperplasia of Schwann cells, axons and connective tissue secondary to traumatic or inflammatory nerve injury. In certain peripheral nerves, traumatic neuromas are identifiable grossly as firm, oval, whitish nodules.⁷ At sectioning, they have a dense fibrous appearance with little vascularity. These nodules appear as tender nodules in the lines of incision 1–10 years after neck dissection. Aside from migraines, patients with traumatic neuromas may also present with paraesthesia distributed over the injured area or hypersensitivity to tactile stimuli, with tenderness on percussion, on pressure and on distortion of surrounding tissues.⁸

Neuromas of the head and neck are likely underdiagnosed and therefore underreported in the literature. While this is the first report of greater auricular nerve (branches of spinal nerves C2, C3) neuromas following endolymphatic mastoid shunt surgery, they have been previously described following parotidectomy and rhytidectomy.^{9–10} These neuromas typically occur near the surgical scar in the region of the posterior border of the sternocleidomastoid muscle.⁷ Meanwhile, neuralgia of the lesser occipital nerve (branch of spinal nerve C3), caused by irritation or injury to the divisions of the occipital nerve, myofascial spasm and focal entrapment of the occipital nerve, is known to be associated with headache, tinnitus, scalp paresthesia, nausea, dizziness and visual disturbances.¹¹ However, outright traumatic neuromas of the lesser occipital nerve are underreported. It is not unlikely for this nerve to be damaged during different types of craniotomy incisions.

Meanwhile, recurrent or resistant vertigo may occur after surgery in patients with Meniere's disease. In a study of 111 patients (82.6% of whom had Meniere's) undergoing surgery for vertigo, 57.7% of patients who received an endolymphatic shunt indicated having disequilibrium.¹² The majority of such cases may be attributable to the irritation of the vestibular nerve, iatrogenic osteogenesis or formation of scar tissue.² However, as our knowledge grows of peripherally generated pain triggers causing centrally mediated symptoms, it is believed that the traumatic neuromas as described were responsible for our patient's migraines and

associated disequilibrium. As shown by the successful diagnostic blockade and eventual resection of the neuromas, the patient did not fail endolymphatic sac surgery for her Meniere's, but rather was experiencing postoperative symptoms due to iatrogenic neuralgia.

Learning points

- In patients with Meniere's and head and neck surgery, attempts should be made to distinguish between treatment failure and the potential development of traumatic neuromas as both may present with migraine-like symptoms.
- Migraine symptoms may include tinnitus, perceived fluctuant hearing, spontaneous spells of vertigo, motion intolerance and motion-provoked vertigo with lightheadedness.¹³
- Such a presentation may fail to raise the suspicion of traumatic neuromas, as migraine headaches have a sixfold higher incidence in patients with Meniere's,¹⁴ and second, migraine-associated dizziness can be indistinguishable from Meniere's disease.
- On the basis of a clinical assessment suggesting a new onset of such symptoms, the formation of traumatic neuromas should be considered.
- Anaesthetic blockade may identify peripheral nerve triggers of chronic headache and are thus both diagnostic and therapeutic.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. **Paparella MM.** Pathogenesis of Meniere's disease and Meniere's syndrome. *Acta Otolaryngol Suppl* 1984;**406**:10–25.
2. **Ghossaini SN, Wazen JJ.** An update on the surgical treatment of Meniere's diseases. *J Am Acad Audiol* 2006;**17**:38–44.
3. **Paparella MM, Fina M.** Endolymphatic sac enhancement: reversal of pathogenesis. *Otolaryngol Clin North Am* 2002;**35**:621–37.
4. **Leibrock LG, Moore GF, Yetter M.** Malpositioned endolymphatic subarachnoid shunt causing trigeminal neuralgia: case report. *Acta Neurochir (Wien)* 1994;**126**:192–4.
5. **Guyuron B, Kriegl J, Davis J, et al.** Comprehensive surgical treatment of migraine headaches. *Plast Reconstr Surg* 2005;**115**:1–9.
6. **Kung TA, Guyuron B, Cederna PS.** Migraine surgery: a plastic surgery solution for refractory migraine headache. *Plast Reconstr Surg* 2011;**127**:181–9.
7. **Lee EJ, Calcaterra TC, Zuckerbraun L.** Traumatic neuromas of the head and neck. *Ear Nose Throat J* 1998;**77**:670–4.
8. **Huang LF, Weissman JL, Fan C.** Traumatic neuroma after neck dissection: CT characteristics in four cases. *AJNR Am J Neuroradiol* 2000;**21**:1676–80.
9. **Moss CE, Johnston CJ, Whear NM.** Amputation neuroma of the great auricular nerve after operations on the parotid gland. *Br J Oral Maxillofac Surg* 2000;**38**:537–8.
10. **de Chalan T, Nahai F.** Amputation neuromas of the great auricular nerve after rhytidectomy. *Ann Plast Surg* 1995;**35**:297–9.
11. **Kuhn WF, Kuhn SC, Gilberstadt H.** Occipital neuralgias: clinical recognition of a complicated headache. A case series and literature review. *J Orofac Pain* 1997;**11**:158–65.
12. **Teufert KB, Berliner KI, De la Cruz A.** Persistent dizziness after surgical treatment of vertigo: an exploratory study of prognostic factors. *Otol Neurotol* 2007;**28**:1056–62.
13. **Shepard NT.** Differentiation of Meniere's disease and migraine-associated dizziness: a review. *J Am Acad Audiol* 2006;**17**:69–80.
14. **Ibekwe TS, Fasunla JA, Ibekwe PU, et al.** Migraine and Meniere's disease: two different phenomena with frequently observed concomitant occurrences. *J Natl Med Assoc* 2008;**100**:334–8.

This pdf has been created automatically from the final edited text and images.

Copyright 2012 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.

BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Please cite this article as follows (you will need to access the article online to obtain the date of publication).

Vorobeichik L, Fallucco MA, Hagan RR. Chronic daily headaches secondary to greater auricular and lesser occipital neuromas following endolymphatic shunt surgery. *BMJ Case Reports* 2012;10.1136/bcr-2012-007189, Published XXX

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow