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Symptomatic Postsurgical Traumatic Neuromas

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Abstract

Hypothesis—Growing nerve fibers from the stumps of amputated sensory nerves can form traumatic neuromas within inner or middle ear postsurgical fibrosis and may produce symptoms commensurate with the normal function of the nerve involved, i.e. balance or pain.

Background—Microscopic traumatic neuromas have been identified in postoperative middle and inner ear fibrosis in the temporal bones of patients complaining of intractable pain or imbalance.

Methods—Postsurgical temporal bones having inner or middle ear traumatic neuromas were reviewed. Twelve of 20 bones with inner ear fibrosis following a variety of neurotologic surgeries were found to have traumatic neuromas, most from the utricular nerve or lateral canal. Five ears in four patients with middle ear fibrosis following chronic ear surgery had traumatic neuromas arising from Jacobson's nerve. An additional 58 bones from chronic ear surgery patients with no neuromas served as a control group. Neurofilament immunohistochemistry labeling substantiated the presence of nerve fibers. Clinical symptoms noted from the clinical records were compared between those with and without traumatic neuromas.

Results—Nine of the 12 patients (75%) with inner ear traumatic neuromas clinically reported constant dysequilibrium postsurgery, lasting for years. None (0%) without neuromas reported new symptoms postoperatively ($p \leq .001$). All five ears with middle ear traumatic neuromas experienced otalgia postsurgery, while none of the other 58 patients with no neuroma reported this problem.

Conclusions—Postoperative intractable disequilibrium or pain may occur as a result of the formation of traumatic neuromas in scar tissue in the inner or middle ears.

Introduction

For years the mechanism by which traumatic and amputation neuromas produced recurrent or persistent pain remained an enigma until Doupe, Cullen and Chance, in 1944, were able to demonstrate ephaptic transmission (false synapse) of efferent autonomic fibers to afferent sensory fibers within the neuromas (1). They first did a local anesthetic block of the sympathetic outflow to the affected limb and, when this produced a temporary cessation of symptoms, performed a sympathectomy that permanently relieved the symptoms. Interaction of neighboring nerve fibers had been demonstrated a year previously by Marrazzi and Lorente de No (2). Histologically, the tumors are represented by disorganized nerve fibers embedded in fibrous tissue (3,4).

Efferent ephaptic transmission of efferent motor impulses to afferent autonomic fibers may also occur as illustrated by gustatory lacrimation due to cross talk between salivary impulses stimulating the lacrimal gland via an injured facial nerve and gustatory sweating due to salivary

impulses affecting the skin of the temple via a damaged auricular temporal nerve that carries both parasympathetic and sympathetic fibers (5).

Here we present evidence that efferent ephaptic transmission may occur in traumatic neuromas in the vestibule and produce prolonged disequilibrium and in the middle ear causing severe disabling otalgia.

Materials and Methods

A search of our database of 808 pairs of temporal bones was made for cases of traumatic neuromas. The temporal bones were from our own pledge program and the National Temporal Bone Registry. The clinical record of each pair of bones was available for review and clinical records and histopathological findings had previously been entered into the database. Institutional Review Board approval for this specific study was not required as approval had already been obtained generically for the temporal bone bank and studies that might ensue from that data (IRB # 06-030). A total of 20 bones with fibrosis in the vestibule were found in the database, of which 12 had traumatic neuromas and the other eight served as a control group. Five bones from four patients were found that had traumatic neuromas of the middle ear following chronic ear surgery. There were a total of 58 additional bones in the database from patients having had mastoidectomies but with no traumatic neuromas, and these served as a control group.

The bones had been removed intracranially within 6 to 36 hours of death, fixed in 10% buffered formalin, decalcified in ethylenediaminetetraacetic acid (EDTA), embedded in celloidin, cut into 20 micron sections that were stained with hematoxylin and eosin and mounted on glass slides. Neurofilament immunohistochemistry labeling was used to substantiate the presence of nerve fibers.

Symptom records were compared between those with and without traumatic neuromas. A test for the difference between two binomial distributions (proportions) was performed to compare the labyrinthine fibrosis groups, while Fisher's Exact test was used to compare the mastoidectomy groups. Criterion for statistical significance was set at $p \leq .05$, two-tailed.

Results

The 12 inner ear traumatic neuromas arose from various nerves in the vestibule (Table 1) (Figs 1-4). Nine of the 12 patients suffered from a balance disturbance from the onset of the problem until their death, a period of 2 to 22 years (mean = 7.5 years). Three of these had undergone labyrinthectomy to control vertigo. All three were relieved of their Ménière's attacks but suffered from chronic disequilibrium, one case requiring a wheel chair, until death (Table 1). None (0%) of the control group of eight ears with labyrinthine fibrosis and no neuroma formation had significant dizziness during a follow-up period of 2 to 29 years (mean = 7 years) ($p \leq .001$) (Table 2). Two of these patients had also undergone labyrinthectomy, the same percentage as in the traumatic neuroma group, but reported no continuing dizziness problem.

The five bones from four patients having had chronic ear surgery were found to have traumatic neuromas on the superior segment of Jacobson's nerve (Table 3) (Fig 5). In each case, severe pain deep in the ear occurred several months after radical mastoidectomies. Re-exploration and revision surgery gave temporary relief of a few months. One ear (#630) was revised 33 times over a period of 51 years. The hypertrophic scarred middle ear mucosa was removed at each surgery and there was relief of pain for 2 to 3 months. On one occasion, the introduction of xylocaine into the middle ear gave pain relief for several hours. One patient, case #645, died of medication overdose, possibly a suicide. Three of these cases were recently described elsewhere (6). None of the 58 patients who had undergone mastoidectomies with no evidence

of traumatic neuromas had persistent pain. The difference in presence of pain between these 58 (0%) and the five ears with a traumatic neuroma (100%) was statistically significant in spite of the very small number of subjects in the neuroma group ($p \leq .05$).

The labyrinthine and middle ear cases with traumatic neuromas had in common obliterative fibrosis, of the vestibular or middle ear space, into which nerve fibers grew and formed neuromas.

Discussion

Failure to compensate for the unilateral loss of function of a labyrinth after one year is unusual (7). Nine of 12 individuals presented in this paper with traumatic neuromas found in their temporal bones experienced a balance disturbance from the time of the unilateral vestibular insult until their death from 2 to 22 years later. They all had traumatic neuromas emanating from a vestibular nerve stump in the vestibule, while none of the patients without a neuroma experienced this problem. All five ears with middle ear neuromas on Jacobson's nerve experienced postoperative otalgia, while none of 58 other patients who also underwent chronic ear surgery but whose temporal bones showed no neuroma reported such postoperative ear pain.

Gardner described "cross talk" between efferent and afferent fibers and afferent autonomic impulses as seen in gustatory lacrimation or sweating when the efferent impulses to the salivary glands excite the lacrimal glands or sweat glands of the temporal area (5). It seems logical to assume that efferent outflow in the vestibular nerves might produce afferent impulses, resulting in stimulation of the vestibular nuclei as the nerves contain efferent as well as afferent fibers. The efferent fibers have a bilateral origin in the brain stem and travel to the vestibular organs via the vestibular nerve (8) so that stimulation of the vestibular system on one side can produce an efferent and reflex ephaptic afferent nerve excitation on the opposite side.

Marrazzi and Lorente de No demonstrated that impulses traveling in a group of fibers may set up new impulses in neighboring fibers while Doupe, et al showed that efferent motor impulses could stimulate afferent pain fibers (1,2). Seltzer and Devor, using rats, showed that this ephaptic transmission occurred when an injured nerve ends in a neuroma (9). Visceral afferent fibers from the tympanic plexus to the geniculate ganglion have been described (10,11).

Jacobson's nerve, also known as the tympanic nerve, is the somatic sensory nerve to the middle ear (10). It is a branch of the glossopharyngeal nerve that participates in the formation of the tympanic plexus and provides sensory innervation within the middle ear to the mucosa of the cavity, pharyngotympanic tube, and mastoid air cells. It also contributes general visceral efferent fibers, which leave the tympanic plexus in the lesser petrosal nerve. The tympanic plexus gets contributions from several cranial nerves including V, VII, IX, and X. We believe that after the formation of a traumatic neuroma in the middle ear, ephaptic transmission ensues and involves afferent pain fibers within cranial nerves V and VII.

Broomfield et al reported that otalgia occurring when the basal electrodes were activated in patients implanted because of hearing loss due to transverse temporal bone fractures was due to leakage of the current through the defect in the cochlear wall to Jacobson's nerve on the promontory (12). Our cases of Jacobson's nerve neuromas suggest afferent pain fibers were being stimulated by efferent impulses in the neuromas. Postoperative persistent pain, lasting years and relieved temporarily by surgery or local anesthesia, is unusual and best explained by the traumatic neuromas found on the nerve ends.

The persistence of disequilibrium or otalgia for years after surgery that produces a fibrous obliteration of the vestibule or middle ear should alert one to the possibility of a traumatic neuroma as the cause of the continuing refractory problem.

We have not discussed treatment for symptoms caused by traumatic neuromas because none of these cases were diagnosed pre mortem and, therefore, we have minimal experience with treatment. Some have suggested intracranial sectioning of nerves (e.g. greater superficial petrosal nerves, nervous intermedius) for otalgia, but we hesitate to suggest an intracranial procedure based on speculation and without more definitive evidence. There are anecdotal reports of treating severe otalgia with anticonvulsive therapy, with some success. But we have no personal experience with use of such drugs. Causalgia, or ephaptic transmission in extremities, has been treated by sympathectomy to remove the efferent loop, but this is not possible in cases like those presented here. For dizziness or imbalance due to intralabyrinthine traumatic neuroma, it is likely that vestibular nerve section would relieve the balance disturbance.

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Fig 1.

Case #90. Traumatic neuroma emanating from the utricular nerve in a woman who died at age 75. Dizziness came on gradually following revision of stapedectomy with wire and gel foam. Wire had dislocated into vestibule. Balance disturbance lasted the four years of follow-up. Hematoxylin and Eosin (H&E $\times 100$).

Fig. 2.
Case #393. Traumatic neuroma on the utricular nerve in a patient who died at age 55, fourteen years after a labyrinthectomy for Ménière's disease. Although she no longer had attacks of vertigo, she continued to have dizziness until death. (H&E $\times 40$)

Fig 3.

Case #619. A 61-year-old female whose otosclerosis had been treated with fenestrations. She developed pneumonia and meningitis 34 years later and a bilateral hearing loss. The hearing returned in the left ear only. She developed a balance disturbance that lasted until her death 4 years later. On one occasion had a serious fall due to imbalance. (H&E $\times 40$)

Fig. 4.

Case #640. Traumatic neuroma on the lateral semicircular canal nerve of a 77-year-old woman who developed dizziness and, because of a fall 6 weeks after a cochlear implant, used a walker until her death from lung carcinoma 3 years after implant surgery. (H&E $\times 100$)

Fig. 5.
Case #630. A 76-year-old female who underwent multiple surgeries in both ears for recurrent pain following radical mastoidectomies for chronic otitis media. Revision surgeries or topical xylocaine gave temporary relief. (H&E $\times 100$)

Table I

Labyrinthine fibrosis with traumatic neuroma.

Case #	Age Death	Sex	Etiology	Neuroma Source nerve	Symptoms Pre op	Symptoms Post op	Duration years
90	75	F	Stapedectomy dead ear	Utricular	None	dizzy ataxia	4
132	77	F	Labyrinthectomy	Utricular	Ménière's attacks	Constant disequilibrium	10
180	83	F	Stapes mobilization	Lateral canal	None	Constant disequilibrium	5
206	62	F	Stapedectomy dead ear	Posterior canal	None	None	
261	55	M	Stapedectomy dead ear	Superior canal	None	Constant disequilibrium	4
272	70	F	Labyrinthectomy	Utricular nerve	Ménière's attacks	Constant disequilibrium	5
393	55	F	Labyrinthectomy	Utricular	Ménière's attacks	Constant disequilibrium	11
554	59	F	Implant	Lateral canal	None	None	
619	61	F	Meningitis	Utricular nerve	None	Constant disequilibrium	4
632	42	M	Implant	Lateral canal	None	None	
640	77	F	Implant	Lateral canal	Unsteady	Constant disequilibrium	3
676	85	M	Stapedectomy	Saccular nerve	None	Constant disequilibrium	22

Table 2

Labyrinthine fibrosis without traumatic neuroma.

Case #	Age	Sex	Etiology	Symptoms Pre op	Symptoms Post op	Follow-up (yrs)
421 R	54	M	Fracture meningitis	Poor balance	Same	29
421 L	54		Implant	Poor balance	Same	13
459	83	M	Stapedectomy	None	?	?
523	82	F	Labyrinthectomy	None	None	2
540	69	M	Transigmoid Cholesteatoma	None	None	3
577	87	F	Labyrinthectomy	None	None	8
617	71	M	Implant	None	None	16
629	95	F	Stapedectomy	None	None	5

Table 3

Postoperative middle ear fibrosis with Jacobson nerve neuroma

Case #	Age Death	Sex	Number Surgeries	Neuroma Source	Symptoms Pre Op	Symptoms Post Op	Duration (yrs)
523	82	F	7	Jacobson N	Otorrhea	Otalgia	40
534	73	F	1	Jacobson N	Otosclerosis	Otalgia	9
630 R	76	F	33	Jacobson N	Otorrhea	Otalgia	51
630 L	76	F	19	Jacobson N	Otorrhea	Otalgia	51
645	57	F	3	Jacobson N	Otorrhea	Otalgia	32