A case of simultaneous retroauricular traumatic neuroma and arteriovenous aneurysm

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Abstract

Traumatic neuroma is a well-defined clinicopathologic entity that is seldom associated with vascular neoformations. We describe the case of a patient with a postsurgical retroauricular neuroma that was associated with a coexisting arteriovenous aneurysm. We also performed an extensive immunohistologic characterization of the traumatic neuroma.

Introduction

Traumatic neuroma was first described in 1811 by Odier as an enlargement or swelling that developed at the distal end of the proximal segment of the peripheral nerve after a partial or complete cut.[1] This lesion is primarily made up of proliferated endoneural and perineural connective tissue, neurilemma cells, and regenerating neuraxes; it is not neoplastic in origin.[2] It has been generally believed that neuromas are always incited by trauma, even though in some cases no previous injury had been documented.[3] Traumatic neuroma is most likely caused by an injury to the peripheral sensory nerves.[2] Following the trauma, the interruption in the continuity of a nerve results in wallerian degeneration, characterized by the loss of axons in the proximal stump and retraction of axons in the distal segment.[4] The central stump axons in the distal end of the proximal segment begin to sprout by budding, and then they zigzag in all directions.[1] In traumatic neuromas, the proximal ends acquire a cap of fibroblasts that proceed to cicatrization. This fibrosis serves as a barrier around the injured nerve end and provides it physical and chemical protection, but the fibrosis also prevents the regrowing axons from reaching their distal counterparts.[3]

Traumatic neuromas of the head and neck have been reported in the oral cavity, the maxillary division of the trigeminal nerve, the inferior alveolar nerve of the mandible, the auriculotemporal nerve, the glossopharyngeal nerve, and the facial nerve.[5]

We report a case of traumatic retroauricular neuroma that was associated with a coexisting arteriovenous aneurysm. To our knowledge, ours is the first report in the literature of such an association.

Case report

A 21-year-old man presented with a right retroauricular mass at the site of a previous surgical incision. Six years earlier, he had undergone removal of an unspecified benign lesion from the site. On otolaryngologic examination, the new mass measured 2 × 1 cm and was mobile. The overlying skin appeared to be normal. The mass was excised with the patient under local anesthesia. During surgery, the neoplasm was noted to have an angiomatous aspect.

Histologic examination revealed that the mass was made up of two separate and adjacent lesions: (1) a traumatic neuroma that was characterized by distorted neural structures embedded in fibrous tissue and (2) an arteriovenous aneurysm that featured a predominance of arteriole-like vessels (figure 1). Immunohistochemical evaluation of the neuromatous component revealed positivity for S-100 protein (figure 2, A), neurofilaments, neuron-specific enolase, CD56, CD57, and synaptophysin. The vascular component showed strong positivity for smooth-muscle actin (figure 2, B).

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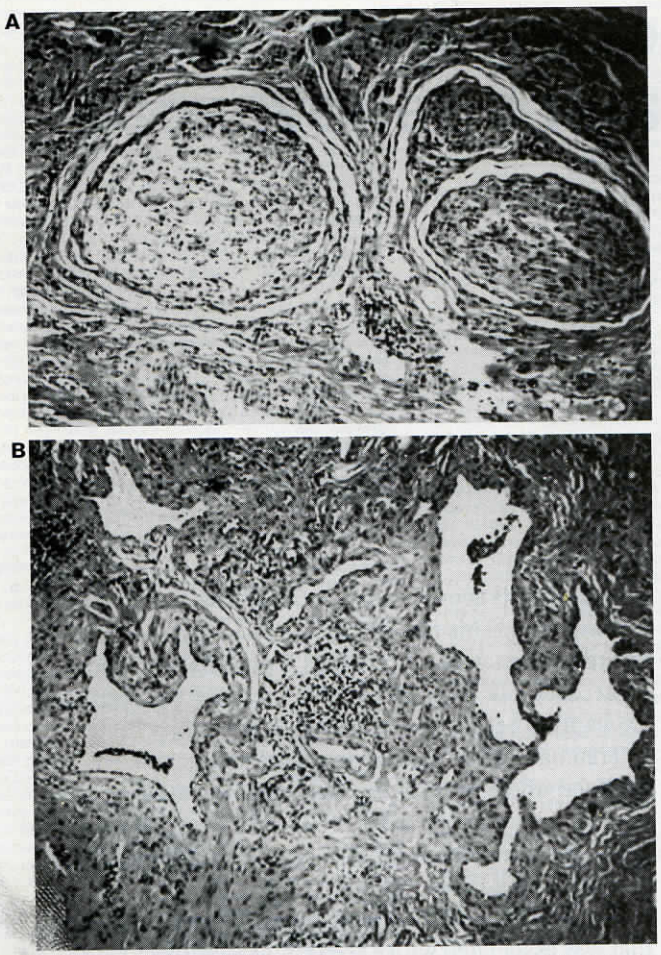

Figure 1. ***A:*** *Two distorted neural structures are evident in the fibrous neuroma tissue.* ***B:*** *Four irregular vascular lumina are seen in this section (both images H&E, original magnification × 25).*

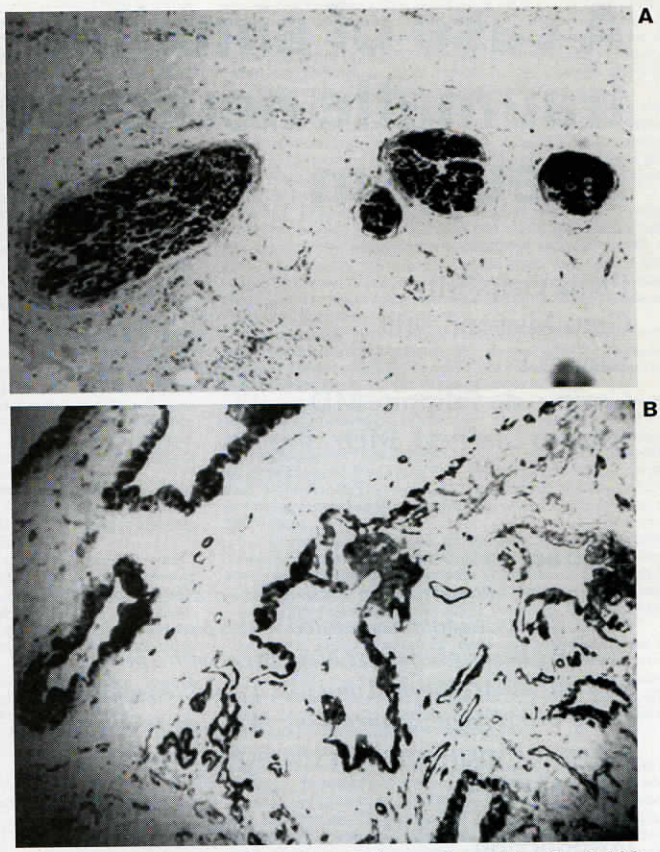

Figure 2. ***A:*** *The neural structure is intensely positive for S-100 protein (immunoperoxidase anti-S-100 protein antibody, original magnification × 20).* ***B:*** *Staining delineates the muscular walls of the aneurysmal component (immunoperoxidase anti-smooth-muscle actin antibody, original magnification × 25).*

Discussion

We performed an extensive immunohistochemical study of the traumatic neuroma. The surgical specimen was tested for S-100 protein, neurofilaments, neuron-specific enolase, CD56, CD57, and synaptophysin (table).

S-100 protein has been demonstrated in both normal and benign neoplastic Schwann's cells; it is positive in virtually all neurofibromas and schwannomas.[6] Immunostaining for neurofilaments and synaptophysin demonstrates the strong positivity of nerve fibers, whereas the perineural epithelial sheaths do not stain.[7,8] Neuron-specific enolase is an enzyme found in the cytoplasm of neurons and cells of neuroendocrine differentiation.[9]

CD56 and CD57 structures are probably involved in cell-cell adhesion in neural tissue. CD56 is identified with an isoform of the nerve-cell adhesion molecule (N-CAM), whereas CD57 detects an epitope expressed on a portion of N-CAM isoforms.[10]

Normally a traumatic neuroma has a dense fibrous appearance with little vascularity.[2] LeMay et al described a hypervascular acoustic neuroma with severe arterio-

Table. Immunohistochemical testing methods

Antibody	Dilution	Source
S-100	1:200	DAKO; Glostrup, Denmark
Neurofilaments	1:50	Immunotech; Milan, Italy
Neuron-specific enolase	1:100	DAKO; Glostrup
CD56	1:50	Novocastra; Newcastle, U.K.
CD57	1:20	Becton Dickinson; Milan
Synaptophysin	1:10	Menarini; Florence, Italy
Smooth-muscle actin	1:50	DAKO; Glostrup

venous shunting.[11] To the best of our knowledge, an association between traumatic neuroma and a vascular lesion has not been previously reported in the literature.

To explain the coexistence of the two lesions, we could hypothesize that there was no association between them and that their proximity occurred only by chance. Another theory is that the arteriovenous aneurysm might have been part of the resected lesion, and the traumatic neuroma might have been the consequence of the incomplete surgical excision. In the latter and more suggestive hypothesis, neuroma formation was stimulated by surgical trauma; the damaged nerve fibers secrete nerve growth factor, which is known to have an angiogenic role[12,13] and might be responsible for aneurysm development.

The treatment of choice for traumatic neuroma is surgical excision. Embedding the cut end of the nerve in soft tissue away from the scar is recommended not only to prevent neuroma recurrence, but to avoid the risk of vascular growth caused by angiogenic factors released by the neuroma.

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