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ISOLATED ACQUIRED PRIMARY GUSTO-LACRIMAL REFLEX FROM A BRAINSTEM GLIOMA

The gusto-lacrimal reflex, also known as Bogorad syndrome, is an autonomic synkinesia in which patients tear excessively in response to salivary stimuli.^{1,2} It most often results from aberrant reinnervation following acute idiopathic or traumatic facial nerve palsy. In rare cases, it occurs as a primary phenomenon in the setting of a slow-growing lesion, such as a vestibular schwannoma or meningioma in the internal auditory canal, that compresses or infiltrates the nerve.^{3,4} We describe a patient with gusto-lacrimal reflex without other evidence of facial nerve dysfunction from a low-grade glioma infiltrating the brainstem and cerebellum.

Case report. A 19-year-old man with an unremarkable medical history was healthy until 6 years earlier when he began to experience occasional frontal headaches, usually in the evening, unassociated with nausea or vomiting but occasionally with photophobia. Three years later, the patient and his parents noted that his right eye would tear while he was eating. The tearing was unassociated with redness of the eye or eye pain. The patient was referred to the Neuro-Ophthalmology Division of the Wilmer Eye Institute, the Johns Hopkins Hospital, for an assessment.

On examination, the patient was alert and cooperative. Best-corrected visual acuity, color vision, and visual fields were normal. There was mild anisocoria, but both pupils reacted normally to light and near stimulation, and there was no relative afferent defect in either eye. Extraocular movements were full. The patient had a small esophoria in primary position and in all positions of gaze but had normal stereopsis. In primary position, he had a fine, right-beating nystagmus that increased in both frequency and amplitude in right gaze and became slightly left beating in left gaze. Saccades from right to center were accurate; however, on saccades from left to center, there was either rebound nystagmus or a resetting of the nystagmus to right beating. Corneal and facial sensation were equal and normal bilaterally. Facial movement was normal. Specifically, there was no evidence of any facial weakness on the right side. The eyelids were in normal position; there was no ptosis or lid retraction. The rest of the examination, including slit-lamp biomicroscopy, intraocular pressure measurements, and ophthalmoscopy, revealed

no abnormalities. A diagnosis of primary gustatory lacrimation associated with central vestibular nystagmus was made, and the patient underwent MRI that revealed an infiltrating enhancing lesion beginning superiorly at the level of the right brachium pontis (figure, A and C), extending inferiorly to involve the right inferior cerebellar peduncle and lateral medulla.

The patient subsequently underwent biopsy of the lesion with removal of its exophytic component. The final pathology was consistent with a low-grade pilocytic astrocytoma.

Discussion. The superior salivatory nucleus is located in the pontine tegmentum just caudal to the motor nucleus of the facial nerve (figure, B and D). It contains the parasympathetic and somatosensory neurons of the facial nerve. These fibers travel to the geniculate ganglion as the nervus intermedius, where they divide into 2 branches: the chorda tympani that provides taste to the anterior portion of the tongue and the greater superficial petrosal nerve that innervates the lacrimal, nasal, and palatine glands.

The gusto-lacrimal reflex is a well-recognized sequela of peripheral facial nerve palsy. The most likely mechanism is misdirection of regenerating gustatory fibers destined for the salivary glands, so that they become secretory fibers to the lacrimal gland and cause homolateral tearing while the patient is eating.⁵ Another hypothesized explanation for this phenomenon is ephaptic transmission and reorganization within the superior salivatory nucleus.⁶ Although the gusto-lacrimal reflex occurs most often after an acute facial nerve paresis and may or may not be associated with other evidence of facial nerve dysfunction,² slow-growing lesions that affect the facial nerve rarely can cause a primary gusto-lacrimal reflex.^{3,4} In such cases, other evidence of facial nerve dysfunction, including aberrant regeneration of the motor nerves, almost always is present. A few cases of congenital gusto-lacrimal reflex with no other signs of facial nerve dysfunction have been reported, but most have been associated with other cranial neuropathies, most commonly abducens nerve paresis, suggesting a developmental abnormality in the pontine tegmentum.⁷ Our case is unique in that the patient's only evidence of facial nerve dysfunction was an acquired gusto-lacrimal reflex. We postulate

(A) Coronal, T1-weighted, postcontrast MRI reveals the rostral and caudal borders of the tumor as well as involvement of the cerebellar peduncle. (B) Artist's drawing shows corresponding location of superior salivatory nucleus. (C) Sagittal, T1-weighted, postcontrast MRI shows that the lesion involves the right pontine tegmentum, the dorsal lateral medulla, and the brachium pontis. (D) Artist's drawing shows corresponding location of the superior salivatory nucleus, which appears to be involved by the lesion.

that this phenomenon resulted from selective damage to the superior salivatory nucleus, as the imaging indicates that the lesion clearly involves the pontine tegmentum where this nucleus is located (figure, B and D).

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