GLOSSOPHARYNGEAL NEURALGIA AND OSSIFICATION OF THE STYLOHYOID LIGAMENT

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The etiology of glossopharyngeal tic, like that of trigeminal neuralgia, has remained an enigma. Despite the rarity of tic douloureux of the glossopharyngeal nerve (compared to trigeminal neuralgia: 1–75), a relatively large number of reports of the former, considered to be unusual, suggest that the cause may be a varied one.

Pudenz and Shelden\textsuperscript{13} described typical glossopharyngeal tic in a patient who had had a shrapnel wound of the neck. Other unusual cases have been reported in which the tic was caused by vascular lesions (vertebral artery atheroma, thrombosis, aneurysm and anomaly),\textsuperscript{2,7,12} and new growth of the cerebellopontine angle.\textsuperscript{3,20} Dandy\textsuperscript{3} was impressed that the incidence of tumor causing glossopharyngeal tic was relatively higher than that of tumor causing trigeminal tic and stressed that cerebellopontine-angle and nasopharyngeal tumor should be sought in patients with a diagnosis of glossopharyngeal neuralgia. He found no difference in the histories of patients with glossopharyngeal tic caused by tumor and those in whom the pain was “idiopathic.” In a series of 20 patients collected from the literature, he noted a known incidence of tumor in 15 per cent and a probable one in 25 per cent.

Glossopharyngeal tic was so named by Harris,\textsuperscript{6} who formulated the idea of “tic pain” from observing a patient who had an epithelioma in the tonsil with similar paroxysmal pain. Syncope, convulsive activity, and cardiac arrest, associated at times with glossopharyngeal neuralgia, emphasize the unusual character of the disease.\textsuperscript{3,14–17,19,21}

Interest in the present problem was stimulated by a patient with left glossopharyngeal tic and unusual clinical features which included ipsilateral ossification of the stylohyoid ligament. Because of the anatomical proximity of the glossopharyngeal nerve to this structure, a relationship was thought possible. Paroxysms of tic pain were referred to the left side of the throat and were precipitated by swallowing, turning of the head to the left side, and mild pressure applied to the left lateral aspect of the hyoid bone. Limitation in flexion and lateral deviation of the neck to the left side was noted. Roentgenograms of the cervical region revealed an ossified structure between the left styloid process of the temporal and lesser cornu of the hyoid bones (Fig. 1). The patient was operated upon on March 26, 1956 and has been free of pain since removal of the mass. Dr. Giancarlo Piazza,\textsuperscript{11} concerned
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with the care of this patient, permitted review of the records of a similar patient relieved of pain in the same way.

HISTORICAL BACKGROUND

The first recorded instance of ossification of the stylohyoid ligament was made by de Marchettis\textsuperscript{10} in a monograph on anatomy published in Padua in 1652. Lücke\textsuperscript{9} (1870) described the cases of 2 women, 20 and 30 years of age, who had difficulty in swallowing because of a long stylohyoid process "pressing the tonsil inward" in one, narrowing the posterior nares on the affected side. Dwight\textsuperscript{4} in an anatomical study described 19 cases of ossification of the stylohyoid ligament. In 10 cases it was bilateral; in 5, on the right side; in 1, on the left. There was no comment in the other 3. Nine were found in men; 3 in women. Six subjects were under 30 and 6 over 50 years of age. Dwight rejected the term "ossification of the stylohyoid ligament" as wrong and misleading, and was of the opinion that the condition was caused by continued growth and subsequent ossification of the second branchial cartilage. Ossification of the second branchial (Reichert's) cartilage, subsequently represented by the styloid process, the stylohyoid ligament and the lesser cornu of the hyoid bone, is not rare, segmental ossification of the chain being observed frequently in the dissecting laboratory. Lipshutz,\textsuperscript{8} having observed a patient complaining of a "sensation of drag-
ging or cutting in the throat,” which was increased by swallowing, made and confirmed by roentgen ray the diagnosis of ossification of the stylohyoid ligament, after palpation of the lateral pharyngeal wall and tonsil revealed a hard mobile mass that appeared fixed to the hyoid bone. General reference was made to treatment by fracture and removal of part of the ossified mass. His case was almost unique in that there was only one recorded previously in which no motion between segments of the chain was demonstrated. He referred to Glazebrook’s case (1908) of complete ossification of the ligament, found in a suicide. There was no comment about symptomatology.

ANATOMICAL CONSIDERATIONS IN THE PRODUCTION OF GLOS SOPHARYNGEAL PAIN (Fig. 2)

According to Gray,⁵ the glossopharyngeal nerve “at its exit from the skull, passes forward between the internal jugular vein and internal carotid artery; it descends in front of the latter vessel, and beneath the styloid process and the muscles connected with it, to the lower border of the stylopharyngeus. It then curves forward, forming an arch on the side of the neck and lying upon the stylopharyngeus and constrictor pharyngis medius. Thence it passes under cover of the hyoglossus, and is finally distributed to the palatine tonsil, the mucous membrane of the fauces and base of the tongue, and the mucous glands of the mouth.”

The stylohyoid ligament, ordinarily a tough band of fibrous tissue, 3–4 mm. wide and 1 mm. thick, lies in immediate lateral relation to the glossopharyngeal nerve, which supplies the stylopharyngeus muscle and to which it is “fixed” by several branches. In the ossified state, the ligament has assumed large proportions so that it may measure 1 cm. or more in
thicknes. The stylopharyngeus draws the side of the pharynx upward and laterally, the ligament being drawn superiorly. When such a fixed bony stylohyoid strul is present, the nerve, during the act of swallowing, is pulled against it and may be stimulated mechanically to produce the painful paroxysm.

**INCIDENCE OF STYLOHYOID LIGAMENT OSSIFICATION IN PATIENTS WITH NINTH NERVE TIC**

The records of 12 patients (6 men and 6 women) with glossopharyngeal neuralgia were reviewed. Eleven were culled from the files of the Buffalo General Hospital; the source of the other was mentioned above. Ten of these 12 patients were operated upon. In 8, the ninth nerve was sectioned intracranially; in the other 2 treatment consisted in the removal of an ossified stylohyoid ligament. In a follow-up of these patients, 8 were re-examined. Two of the original 12 had expired since their intracranial operation; a third patient refused re-examination. Two of 6 patients who had had intracranial operation were found by roentgen ray to have ossified stylohyoid ligaments, 1 bilaterally. Roentgenograms of the neck of a man who was not operated upon revealed a very long styloid process on the affected side with a pseudo-articulation at its base. In the other, the ligament was ossified bilaterally, being fused to the hyoid bone and better developed on the side of the pain. The remaining 2 treated by removal of the ossified ligament are described above. Though this series is too small to be significant statistically, there was an incidence of ossification of the stylohyoid ligament in at least 50 per cent (6 of 12 patients). Since 3 of the patients could not be examined, it is not known whether these patients did in fact have ossification of the ligament. The relative rarity of tic of the ninth nerve and the high incidence of ossification of the ligament in this series appears to represent more than mere coincidence in their relationship. In only 2 of this series of patients was the ossification bilateral. The ossified strut was present in 4 men and 2 women.

Sicard and Robineau18 and Adson1 did 7 operations on the neck in patients with evident glossopharyngeal neuralgia. It would appear that had ossified ligaments been present in their cases, they would have been described.

**DIAGNOSIS**

It appears from this study that typical glossopharyngeal tic may be associated with ossification of the stylohyoid ligament. Physical examination reveals limitation in flexion of the neck and deviation of the head to the affected side in patients with complete ossification of the ligament. The paroxysms of pain may be reproduced by swallowing, turning of the head to the affected side, and by pressure applied to the lateral side of the neck at the hyoid bone. A hard mass is palpable in the lateral wall of the pharynx.

The radiological findings are the most significant. Roentgenograms will reveal the ossified mass with or without joint formation. Ossification may
be incomplete, in such cases the chain being represented by nonradiopaque cartilage. It is evident that the incidence of cartilage formation in the ligament in patients with tic of the ninth nerve can be determined only by exploration of the neck.

SURGICAL CONSIDERATIONS

Wide exposure is necessary. The incision should extend from above the mastoid tip, obliquely across the anterior cervical triangle to the midline over the septum of the hyoid bone, the neck being well extended and turned slightly away from the affected side (a small blanket roll placed beneath the shoulders helps in such positioning). Dissection is made gingerly in freeing the ossified pillar, remaining close to it at all times, carefully packing the surrounding medial tissues away with cottonoid. This is particularly important superiorly, where the facial nerve could be injured, where it makes its exist from the stylomastoid foramen. Retraction of the tissues on the upper bank of the incision superiorly should be gentle to avoid injury to the inferior alveolar and lingual branches of the mandibular nerve. It is suggested that the bone be removed piecemeal in at least its upper one-half (rather than to disarticulate it), so as to minimize the chance of injury to the seventh nerve, even though a joint may be present at the base of the styloid process. In its inferior one-half the bone can be safely rocked and twisted from the lesser cornu of the hyoid bone, if it is not completely ossified.

SUMMARY

It is evident that the etiology of glossopharyngeal neuralgia in a relatively high percentage of patients may have a mechanical basis. This is because of direct stimulation of the nerve trunk against an ossified (or perhaps cartilaginous) stylohyoid ligament. Removal of the mass resulted in relief of the patient’s pain in 2 cases. The high incidence of ossified stylohyoid ligament in patients with glossopharyngeal tic is significant. Why spontaneous relief of pain occurs in these patients (as in those with trigeminal neuralgia) is, however, difficult to explain on this basis.

It is suggested that roentgen-ray examination of the necks of all patients with glossopharyngeal tic be made which may help in a better understanding of the problem.

REFERENCES

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