

Short report

Neck tongue syndrome: operative management

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SUMMARY A 53-year-old woman with assimilation of the atlas to the occiput presented with paraesthesiae in the right half of her tongue and ipsilateral neck pain aggravated by head turning. After being intermittent for several years, the symptoms eventually became persistent and increasingly incapacitating. At operation, the C2 spinal nerves were found to be compressed by protuberant atlanto-axial joints, particularly on the right side. The superficial parts of the resected C2 spinal nerves showed a loss of both myelinated and unmyelinated nerve fibres. After operation, the patient experienced partial relief of her symptoms.

Lance and Anthony¹ introduced the term "neck-tongue syndrome" to describe four young patients with transient neck pain and ipsilateral tongue numbness precipitated by head turning. The symptoms of this syndrome were attributed to compression of the C2 ventral ramus by abnormal subluxations of the lateral atlanto-axial joints.² Tongue symptoms are presumably caused by the involvement of afferent fibres from the lingual nerve that pass through the hypoglossal nerve to the C2 ventral ramus.¹ We describe a middle-aged patient who developed persistent unilateral neck pain and tongue numbness. Surgical exploration of the upper neck established that the C2 spinal nerves were stretched across the atlanto-axial joint spaces, particularly on the affected side. The resection of these injured nerves provided partial symptomatic relief.

Case history

A 53-year-old right-handed woman presented in September, 1980 with a 6 year history of pain in the right side

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Received 3 June 1983 and in revised form 23 October 1983.
Accepted 5 November 1983

Presented in part at the 17th Canadian Congress of Neurological Sciences, June, 1982

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of her neck. The discomfort, which was initially intermittent but had become constant in the preceding 6 months, was aggravated by head turning and talking but was not affected by coughing or straining. When severe, the pain radiated to the back of her head or into the right arm and caused mild dysphagia because of a feeling of muscle spasm in her throat. Since 1979, she had noted an intermittent tingling sensation in the right tongue, lower gum, and base of her mouth; this symptom had become constant in the preceding 6 months. She also experienced tingling in both hands, the right greater than the left, that awakened her during the night and occasionally occurred while she was knitting. Physical examination was unremarkable except for mild, non-localised tenderness over the right upper neck. No objective sensory loss could be demonstrated in the areas of paresthesias in her mouth.

Cervical spine radiographs showed assimilation of the atlas to the occiput; films taken in flexion and extension did not show abnormal movement of the C1-C2 junction. Computed tomography of the head to the C1 vertebral level and complete myelography showed no structural abnormalities. Nerve conduction studies suggested a mild right carpal tunnel syndrome; sensory conduction was 44.7 m/s (amplitude 17 μ V) on the right and 52.6 m/s (amplitude 25 μ V) on the left. No other nerve conduction or electrophysiological abnormalities were detected.

Because the patient's symptoms did not respond to immobilisation with a cervical collar or various medications for pain, the spinal nerves of the upper cervical region were explored surgically in November, 1981. The atlanto-axial interspace measured 35 mm between the neural arches with the patient's neck in a position of partial flexion. The right atlanto-axial articulating process was more protuberant than the left, stretching the right C2 spinal nerve and its rami in a caudal direction so that the ventral ramus joined the dorsal ramus from an anterior direction and passed directly over the capsule of the joint



Fig A Longitudinal section of right C2 ventral ramus. The fibre loss and scarring are more severe near the surface (top); myelinated fibres (arrow) are more numerous deeper in the nerve. (H and E $\times 145$).

space. These nerves were tougher to palpation on the right than on the left. The C2 spinal nerves together with their dorsal and ventral rami were resected bilaterally.

During the first week after operation, the patient experienced no tingling sensations in her tongue but thereafter she reported a recurrence of the sensation in a mild form when she was tired. There was hypaesthesia to pin in the distribution of both greater occipital nerves. For several months post-operatively she required codeine and diazepam for daily recurrent neck pain. After several months, the neck pain markedly subsided but tended to recur with fatigue.

Pathology

The C2 spinal rami, especially the ventral rami, showed nerve fibre loss and fibrosis. Such changes were more severe in the superficial parts of the nerves (fig A). Electron microscopy of these areas showed severe loss of both



Fig B Electron micrograph of cross section of superficial part of ventral ramus. Both myelinated and unmyelinated nerve fibres are replaced by collagen containing few fibrocytes and Schwann cells devoid of axons. Two large nerve fibres remain; one of these is demyelinated. ($\times 1400$).

myelinated and unmyelinated nerve fibres (fig B). The distal fringe of the left dorsal root ganglion, which was included in the resected material, was relatively well preserved but showed slight fibrosis and cell loss. There was also perivascular fibrosis within the rami and fibro-hyaline thickening of many small blood vessels.

Discussion

Certain features of the syndrome reported in the present patient differed from those in the patients of Lance and Anthony.¹ Their patients first noted transient symptoms in childhood whereas our patient was older with more persistent symptoms. Nevertheless, the descriptive term "neck-tongue syndrome" can be used to describe our patient's symptoms because the essential features are similar for both.

The main features of the "neck-tongue" syndrome, neck pain and lingual paraesthesias,¹ are presumably caused by repeated minor subluxations of the C1-C2 articulating processes with compression and stretch injury of adjacent nerves.² The pathologic changes demonstrated in the nerves resected from our patient support this hypothesis; both the hyaline thickening of the small blood vessels and the loss of myelinated and unmyelinated nerve fibres from the superficial portions of these

nerves could have been caused by repeated physical trauma.

The pain of the neck-tongue syndrome is attributed to chronic trauma to the joint capsule of the lateral articulating process. Paraesthesiae, which can be caused by compression of spinal or peripheral nerves,³ presumably occur because of involvement of afferent fibres from the tongue in the upper cervical nerves. Animal experiments and human studies (see ref 1 for review) have demonstrated that such proprioceptive fibres originate within the tongue, are carried initially by the lingual branch of the trigeminal nerve, pass through communicating branches into the hypoglossal nerve, and then join the ventral primary rami of one or more of the upper three cervical spinal nerves to enter the central nervous system through the respective dorsal roots.

Although it has been shown in previous dissections that the ventral primary ramus is most vulnerable to this type of injury because of its usual transverse course across the joint space,² greater degrees of subluxation could also affect the adjacent spinal nerve trunk and the dorsal primary ramus. Such a situation might be expected in patients with atlanto-occipital anomalies in which the articulating functions of the cranio-cervical junction are assumed by the C1-C2 facets. Thus it is probably more than coincidental that two of the four patients previously reported with this syndrome¹ also had atlanto-occipital anomalies.

No completely satisfactory treatment has been reported for the neck-tongue syndrome. Resection of both C2 spinal nerves provided temporary relief for our patient but minor tongue paraesthesias recurred and the neck pain only diminished after several months. The recurrence of the tongue symptoms suggests a facilitation of residual proprioceptive fibres in the adjacent uninjured spinal roots; it is possible that greater amelioration of symptoms would have occurred if the C1 and C3 roots, which may also contain sensory fibres from the tongue,^{4,5} had been resected at the same time as the C2 roots.

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J Neurol Neurosurg Psychiatry 1984 47: 407-409

doi: 10.1136/jnp.47.4.407

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