

An anatomical basis for the Neck–Tongue Syndrome

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SUMMARY The C2 nerve roots and rami were dissected in five cadavers to explore the pathogenesis of Neck–Tongue Syndrome. The most likely cause of the simultaneous occurrence of suboccipital pain and ipsilateral numbness of the tongue is an abnormal subluxation of one lateral atlanto-axial joint with impaction of the C2 ventral ramus against the subluxated articular processes.

Recently, Lance and Anthony¹ described a “Neck–Tongue Syndrome,” affecting four adolescent patients, in whom sudden rotatory movements of the head precipitated unilateral suboccipital pain and ipsilateral numbness of the tongue. The authors argued that the symptoms were due to compression of the second cervical roots in the atlanto-axial space; the numbness of the tongue was caused by compression of proprioceptive fibres coursing from the tongue through the ansa hypoglossi, the cervical plexus, and finally the second cervical dorsal root. To explore the pathogenesis of this syndrome an anatomical study of the C2 spinal nerve, roots and rami was undertaken. This paper reports the results of this study and a more detailed interpretation of the mechanism of Neck–Tongue Syndrome is advanced.

Methods

With the aid of a $\times 40$ dissecting microscope, the C2 spinal nerves, nerve roots and rami were dissected in five embalmed human adult cadavers. To assess the effect on the C2 nerve roots and rami of rotation of the atlas, all muscles attaching to the axis, atlas and skull were resected leaving the nerves in situ. Such preparations allowed the head and atlas to be rotated on the axis through a range of up to 30° to either side and a degree of extension which was limited by contact of the posterior arch of the atlas with the lamina of the axis. The relationship of the nerves in question to the bony elements during these movements could then be studied by direct observation.

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Results

Resecting the dorsal neck muscles does not affect the position and skeletal relations of the extradural C2 roots, spinal nerve and rami. In all ten nerves studied the relations of these nerves were the same.

The C2 ventral and dorsal roots unite to form a short spinal nerve which divides into a dorsal and a ventral ramus. This series of nerves lies dorsal to the lateral atlanto-axial joint (fig 1). The nerves are accompanied by a plexus of veins which communicate between the epidural plexus and the vertebral veins. The nerves and veins are invested by fascia which holds the C2 roots, ganglion and spinal nerve against the capsule of the lateral atlanto-axial joint. The C2 ventral ramus (fig 1) courses transversely across the lateral atlanto-axial joint and along its course gives off from its ventral surface, articular branches to the joint (fig 2). Ultimately it arches over the vertebral artery and enters the cervical plexus. Often the C2 ventral ramus crosses the joint opposite the superior articular process of the axis and is held against the lower half of the capsule of the joint by fascia. Less frequently the ramus may follow a more rostral course and is related to the inferior articular process of the atlas.

During rotation of the head the C2 roots move in company with the articular process of the atlas because they are held against the upper half of the capsule of the lateral atlanto-axial joint by fascia. In rotation to the left, for example, the right atlanteal articular process subluxates ventrally over the superior articular process of the axis. The right C2 roots therefore also move

ventrally in company with the atlas. If the C2 ventral ramus is related to the atlanteal articular process it too displaces ventrally with the atlas. However, when the ventral ramus crosses the axial process the ventral displacement of the atlas causes the ventral ramus to be drawn over the margin of the lateral atlanto-axial joint since distally and laterally the ventral ramus is relatively fixed by fascia to the lower half of the joint capsule.

During normal rotation of the atlas there is a considerable degree of subluxation of the lateral atlanto-axial joint and so, although covered by the joint capsule, the margins of the articular processes forming the joint overlap and form

projecting edges. In rotation to the left, for example, the right atlanteal process displaces ventrally exposing the dorsal margin of the right superior articular process of the axis. In those cases where the right C2 ventral ramus crosses the joint margin (fig 3A), during rotation it is in effect drawn across this exposed dorsal margin, separated from it only by the capsule of the joint (fig 3B). In rotation to the right a similar but inverted effect occurs. The right inferior articular process of the atlas subluxates dorsally, and the right C2 spinal nerve and ventral ramus are drawn across the projecting dorsal edge of the inferior articular process of the atlas (fig 3C).

The C2 roots lie mainly within the vertebral

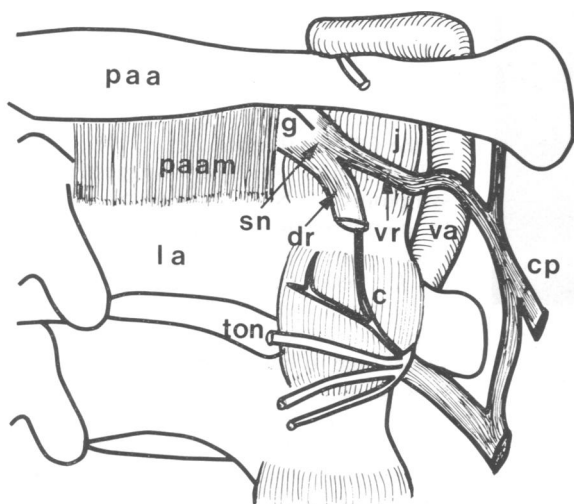
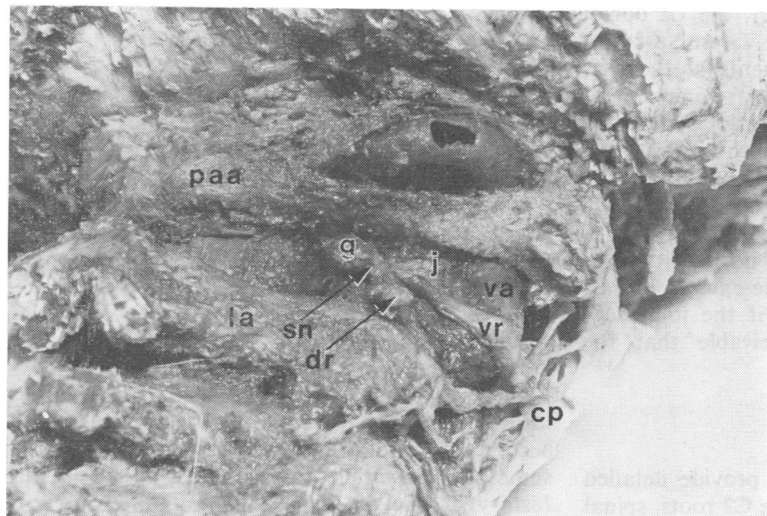


Fig 1 The relationship between the C2 dorsal root ganglion (g), spinal nerve (sn), and ventral ramus (vr) and the lateral atlanto-axial joint (j), posterior arch of the atlas (paa), the lamina of the axis (la), and the vertebral artery (va).

A: Schematic illustrations of a dorsal view. B: Dissection of a right dorsolateral view. dr=C2 dorsal ramus (cut at its point of branching). cp=cervical plexus. ton=third occipital nerve. c=communicating branch C2 to C3. paam=posterior atlanto-axial membrane.



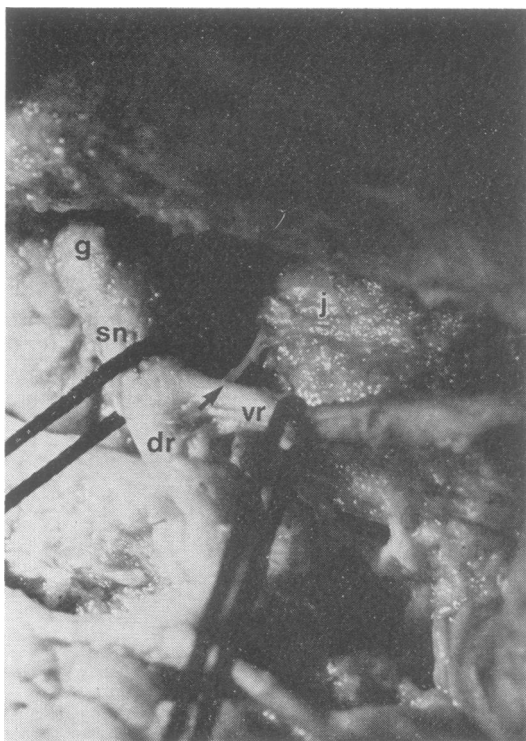


Fig 2 Close up view of an articular branch (arrow) to the left lateral atlanto-axial joint (j) from the C2 ventral ramus (vr). The ramus, spinal nerve (sn) and ganglion (g) have been retracted to expose the articular nerve. dr=C2 dorsal ramus.

canal deep to the atlas. Therefore, they themselves are not susceptible to bony impingement during any motion of the head. However, the C2 dorsal root ganglion lies dorsal to the lateral atlanto-axial joint. During most movements of the axis this ganglion is free from bony impingement (fig 4A and B) but during rotation combined with forced extension it may be compressed between the posterior arch of the atlas and the superior articular process of the axis (fig 4C). This phenomenon has been discussed previously² and it should be noted that, in a normal joint, in order to compress the ganglion the extension must be forced. One the other hand if the joint subluxates abnormally it is conceivable that the ganglion could be compressed.

Discussion

Textbooks of anatomy^{3,4} do not provide detailed descriptions of the relations of the C2 roots, spinal

nerve, and rami. Previous authors⁵⁻⁸ have rectified certain misconceptions and deficiencies relating to the anatomy of these nerves and the description given in the present study agrees with and incorporates these changes. The most notable features highlighted in this and previous studies⁵⁻⁸ are that the C2 ganglion and spinal nerve lie dorsal to the lateral atlanto-axial joint and that this joint is innervated by the C2 ventral ramus. These facts underlie the following interpretation of the mechanism of Neck-Tongue Syndrome.

In the Neck-Tongue Syndrome, numbness of the tongue and suboccipital pain are triggered by rotation of the head. The site of pain and the precipitating manoeuvre clearly implicate an abnormality at upper cervical levels. The tongue symptoms have been discussed by Lance and Anthony¹ who reviewed the available data on the course of afferent fibres from the tongue; and it seems well established that proprioceptive fibres from the tongue do pass via the ansa hypoglossi to the C2 dorsal root.

Lance and Anthony¹ suggested that all the symptoms of Neck-Tongue Syndrome were due to compression of the C2 nerve roots in the atlanto-axial space. However, as described above, the C2 roots lie deep within the vertebral canal and are not susceptible to bony compression. The only elements of the C2 nerves which are susceptible to bony impingement are the C2 dorsal root ganglion and the C2 ventral ramus. The ganglion may be compressed between the posterior arch of the atlas and the superior articular process of the axis, during combined extension and rotation (fig 4C). C2 ventral rami which cross the margin of the lateral atlanto-axial joint may be stretched over the edge of the atlanteal or axial articular process, during rotation of the atlas (fig 3).

Although the other symptoms of Neck-Tongue Syndrome may be explicable in terms of nerve compression, the pain is not, for it has been demonstrated that experimental compression of spinal nerves⁹ or peripheral nerves¹⁰ produces paraesthesiae but not pain. A more satisfying explanation is that patients with Neck-Tongue Syndrome suffer a temporary abnormal subluxation of their lateral atlanto-axial joint which strains the joint capsule, thus causing pain. Subluxations have not been documented in Neck-Tongue Syndrome since, because of the intermittent nature of the condition, patients have not been examined during an attack. However, that some such instability occurs is suggested by the history of one of Lance and Anthony's patients



Fig 3 *The relationship between the C2 ventral ramus and the articular processes of a right lateral atlanto-axial joint during rotation of the atlas (lateral view). The capsule of the joint has been resected to reveal the edges of the articular processes of the atlas (at) and axis (ax). A: Neutral position. The C2 ventral ramus (vr) crosses the joint space and the dorsal aspect of the superior articular process of the axis (ax). B: Rotation to the left. The C2 ventral ramus (vr) is drawn over the exposed dorsal edge of the superior articular process of the axis (ax) and impacted against it (arrow). C: Rotation to the right. The inferior articular process of the atlas (at) displaces dorsally and impacts the C2 ventral ramus (vr) against its dorsal edge (arrow).*

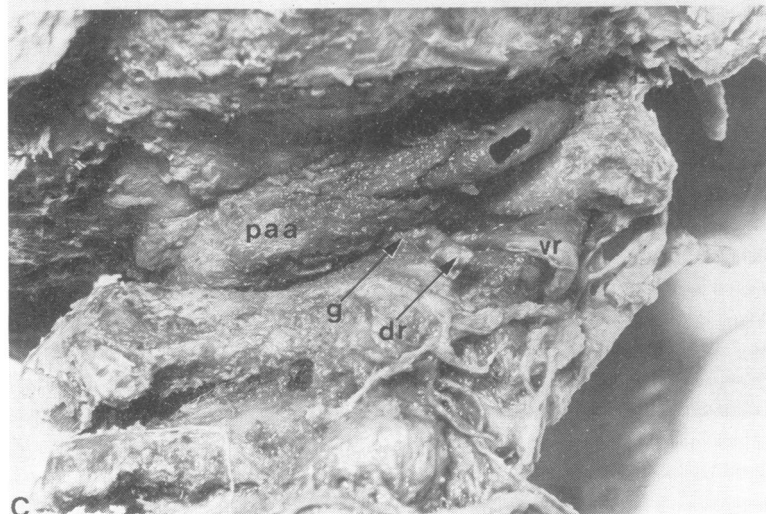


Fig 4 Relationships of the right C2 dorsal root ganglion to the axis and atlas during movements of the atlas. (cf fig 1B for neutral position and key to labelling). A: extension; B: rotation to the right; C: rotation to the left combined with forced extension. Note the compression of the ganglion.

who reported recurrent episodes in which his head "fell back" forcing him to look upwards and to the right. Normal posture was restored by flexion and side to side movement of the head.

Apart from numbness of the tongue, another symptom of Neck-Tongue Syndrome is numbness of the skin behind the ear, reported by two patients.¹ The retro-auricular skin is innervated by the lesser occipital nerve and the posterior branch of the greater auricular nerve both of which are derived from the C2 ventral ramus.³ Proprioceptive fibres from the tongue also return via the cervical plexus to the C2 ventral ramus.¹ Thus all the "numbness" sensations of Neck-Tongue Syndrome are consistent with compression of the C2 ventral ramus. As shown in the present study, during normal rotation of the atlas, the C2 ventral ramus may be impacted against the edge of an articular process of the lateral atlanto-axial joint. If patients with Neck-Tongue Syndrome were indeed suffering in addition an abnormal subluxation of that joint then the likelihood of impaction would be greater.

It may be questioned whether compression or irritation of proprioceptive fibres from the tongue could cause numbness. However, it is well known that patients with Bell's palsy frequently complain of "numbness" although trigeminal sensation is intact.^{11,12} This "numbness" is explicable in terms of compression of the proprioceptive fibres which are known to run in the facial nerve.¹³ Thus the sensation of "numbness" in both Bell's palsy and Neck-Tongue Syndrome does not imply a loss of touch and pain sensation but rather reflects the impaired function of deep proprioceptive afferents.

It is notable that none of the patients described by Lance and Anthony¹ complained of numbness in the paramedian skin of the occiput and vertex. This is the area supplied by the greater occipital nerve which is derived from the C2 dorsal ramus. This sparing of the dorsal ramus fibres indicates that the ventral ramus alone was compressed, and not the dorsal root ganglion, for otherwise symptoms should have occurred in both ventral and dorsal ramus territories. Moreover, the precipitating factor in Neck-Tongue Syndrome is rotation of the head. This manoeuvre may compromise the C2 ventral ramus but not the C2 dorsal root ganglion, which further indicates the exclusive role of the C2 ventral ramus in Neck-Tongue Syndrome.

Notwithstanding this, the vulnerability of the C2 dorsal root ganglion to compression during rotation combined with extension does theoretic-

ally make it a potential source of symptoms, but such a Neck-Tongue Syndrome would then include numbness in the greater occipital nerve territory.

While the present study postulates a plausible mechanism for the principal features of Neck-Tongue Syndrome, it does not explain the numbness of the fingers reported by two patients in Lance and Anthony's series.¹ This symptom cannot be related to C2 ventral ramus compression, and the present data do not provide an explanation. Lance and Anthony¹ felt that it may have been due to traction on the lower nerve roots, exerted through the dura mater, during subluxation of the atlas, but there is no anatomical data to support this hypothesis. Reid¹⁴ studied the extent of movement of the dura and spinal cord during flexion/extension movements of the vertebral column and head, but comparable data with respect to rotation are lacking. Thus an explanation of the upper limb features of Neck-Tongue Syndrome awaits further investigation.

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References

- 1 Lance JW, Anthony M. Neck-tongue syndrome on sudden turning of the head. *J Neurol Neurosurg Psychiatry* 1980; **43**:97-101.
- 2 Bogduk N. The anatomy of occipital neuralgia. *Clin Exp Neurol* 1980 (in press).
- 3 Gray's Anatomy. 35th ed. Warwick R, Williams PL, ed. London: Longmans 1973.
- 4 Cunningham DJ. Textbook of Anatomy. 11th ed. Romanes GJ ed. London: Oxford University Press, 1972.
- 5 Guerrier Y, Colin R. *Le deuxième nerf cervical* CA Ass Anat 41e Congres 1954; 813-6.
- 6 Lazorthes G, Gaubert J. *L'innervation des articulations interapophysaires vertebrales*. CR Ass Anat 43e Reunion 1956; 488-94.
- 7 Juskiewinski S, Lazorthes F, Boulard PY, Lazorthes G. *Le deuxième nerf cervical*. CR Ass Anat 53e Congres 1968; 1044-7.
- 8 Lazorthes G. Les branches posterieures des nerfs rachidiens et le plan articulaire vertebraire posterieur. *Ann Med Phys* 1972; **15**:192-202.
- 9 MacNab I. The mechanism of spondylogenic pain. In: Hirsch C, Zotterman Y, eds. *Cervical Pain*. Oxford: Pergamon 1972; 89-95.
- 10 MacKenzie RA, Burke D, Skuse NF, Lethlean AK. Fibre function and perception during cutaneous nerve block. *J Neurol Neurosurg Psychiatry* 1975; **38**:865-73.

- 11 Davidson's Principles and Practice of Medicine 11th ed. MacLeod J, ed. Edinburgh: Churchill Livingstone 1974; 821.
- 12 Victor M, Adams RD. Diseases of cranial nerves. In Harrison's *Principles of Internal Medicine* 8th ed. Isselbacher KJ, Adams RD, Braunwald E, Petersdorf RG, Wilson JD, eds. New York: McGraw-Hill, 1980; Ch 376; 202–3.
- 13 Fay T. Atypical neuralgia. *Arch Neurol Psychiat* 1927; **18**:309–15.
- 14 Reid JD. Effects of flexion-extension movements of the head and spine upon the spinal cord and nerve roots. *J Neurol Neurosurg Psychiat* 1960; **23**:214–21.