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 Anthony 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 root 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 ×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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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

![Diagram](http://jnnp.bmj.com/)

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).

nerve, and rami. Previous authors\(^5\)\(^\text{-}^8\) 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 studies\(^5\)\(^\text{-}^8\) 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 Anthony\(^1\) 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 Anthony\(^1\) 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 nerves\(^9\) or peripheral nerves\(^10\) 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.

**Discussion**

Textbooks of anatomy\(^3\)\(^\text{-}^4\) do not provide detailed descriptions of the relations of the C2 roots, spinal

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*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.*
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.
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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. 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. Proprioceptive fibres from the tongue also return via the cervical plexus to the C2 ventral ramus. 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 atlantoaxial joint. If patients with Neck–Tongue Syndrome were indeed suffering in addition an abnormal subluxation of that joint then the likelihood of inpa...


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