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The C2-tongue connection: anatomically tasteful but often forgotten

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Statement

We present a case of post-operative numbness of the tongue after surgical resection of an intradural C2 meningioma. This case report is the first to describe an ipsilateral numbness of the tongue after manipulation of the C2 nerve root in spinal meningioma surgery. The underlying explanatory anatomy for the clinical symptoms has previously been described in the neck-tongue syndrome. This anatomical relationship is particularly interesting, but also important for spine surgeons performing complex cervical spine procedures.

Clinical description

A 50-year-old patient was referred to our neurosurgical department with a cerebellopontine angle meningioma. During the diagnostic work-up, a large right-sided intraspinal extramedullary mass at the second cervical level was identified, causing compression of the spinal cord. MRI imaging showed an intraspinal extramedullary retro-corporal mass at the base of the C2 vertebra compatible with a meningioma. (Figure 1)

In consultation with the multidisciplinary surgical team, the spinal meningioma at C2 was treated first. The surgery was performed using intra-operative neuromonitoring using somatosensory evoked potentials (SEPP) and motor evoked potentials (MEP) in collaboration with the neurology department. The intradural mass was visualized after a C2 laminectomy and durotomy. The mass was totally resected (Simpson grade II). The C2 nerve root didn't had to be sacrificed, nor the dentate ligament was cut. SEPP and MEP signals remained uninterrupted during the procedure.

Post-operatively the patient had post-operative nausea and vomiting (PONV) and complained of right-sided numbness of the tongue, auricular pain and irritation of the skin on and behind the ear. The rest of the clinical examination was completely normal. The patient was able to leave the hospital after one week. Histopathological examination confirmed a WHO-grade 1 meningioma.

One month after the surgery, the patient was seen at the neurosurgical outpatient clinic. The numbness of the tongue and auricular pain improved significantly. The residual symptoms consisted of some numbness posteriorly to the ear on her head. The patient was informed that further improvement could be expected over time. A post-operative MRI scan was performed to confirm the extent of the resection. This MRI scan showed no residual meningioma (figure 1).

Discussion and conclusion

We hypothesized that the symptoms could be explained by the anatomical relation between the hypoglossal nerve and the cervical nerve roots. This anatomical correlation was initially mentioned in the literature as neck-tongue syndrome. The neck-tongue syndrome is a rare condition characterized by pain in the upper cervical/occipital areas associated with ipsilateral glossal paresthesia, mostly after sudden neck movement. (1) Anatomical studies have illustrated the association between the cervical plexus and the hypoglossal nerve. Lance and Anthony were the first to describe the anatomical correlation between the hypoglossal nerve and the second cervical nerve root (2). The paired hypoglossal nuclei extends in the dorsal portion of the medulla oblongata. Two bundles of hypoglossal nerve rootlets leave the medulla between the pyramid and the inferior olive. These rootlets form the hypoglossal nerve and exit the base of the skull through the hypoglossal canal. Here it is joined by somatic efferent fibers from the first cervical nerve C1 nerve, which form the upper root of the ansa cervicalis. This loop of nerve tissue is continued in the inferior root

consisting of the descending C2 and C3 somatic efferent nerve fibers. (3) Next, afferent proprioceptive fibers anastomose with the hypoglossal nerve in the tongue. The connection between the upper cervical nerve roots and the hypoglossal nerve, **allows these fibers to continue to the spinal cord through the C2 nerve**. This implies that the numbness of the tongue, could primarily be a manifestation of the loss of touch and proprioception in the afferent nerve fibers that reach the C2 spinal cord. Next, the irritation on the skin of the ear and right behind the ear can be explained by the afferent fibers from the greater auricular nerve and the lesser occipital nerve, which **both descend** to the spinal cord through the C2 nerve roots. (1, 4) The relation between the C2 nerve root and the hypoglossal nerve is schematically presented in figure 2, based on a comprehensive anatomical review from Shoja et al. (5)

The surgical procedure in this case was uncomplicated. During the surgery, the C2 nerve has been manipulated, as its anatomical position made it impossible not to do this. No macroscopic damage to the C2 nerve was objectified at any time, nor direct coagulation was used.

As a conclusion, we hypothesize that surgical manipulation of the C2 nerve should have caused a temporary ipsilateral numbness of the tongue and irritation around the ear, based on the anatomical relation between the hypoglossal nerve, the ansa cervicalis, the C2 root, the greater auricular nerve and the lesser occipital nerve. To our knowledge, no comparable case has been reported in the literature.

Figures:

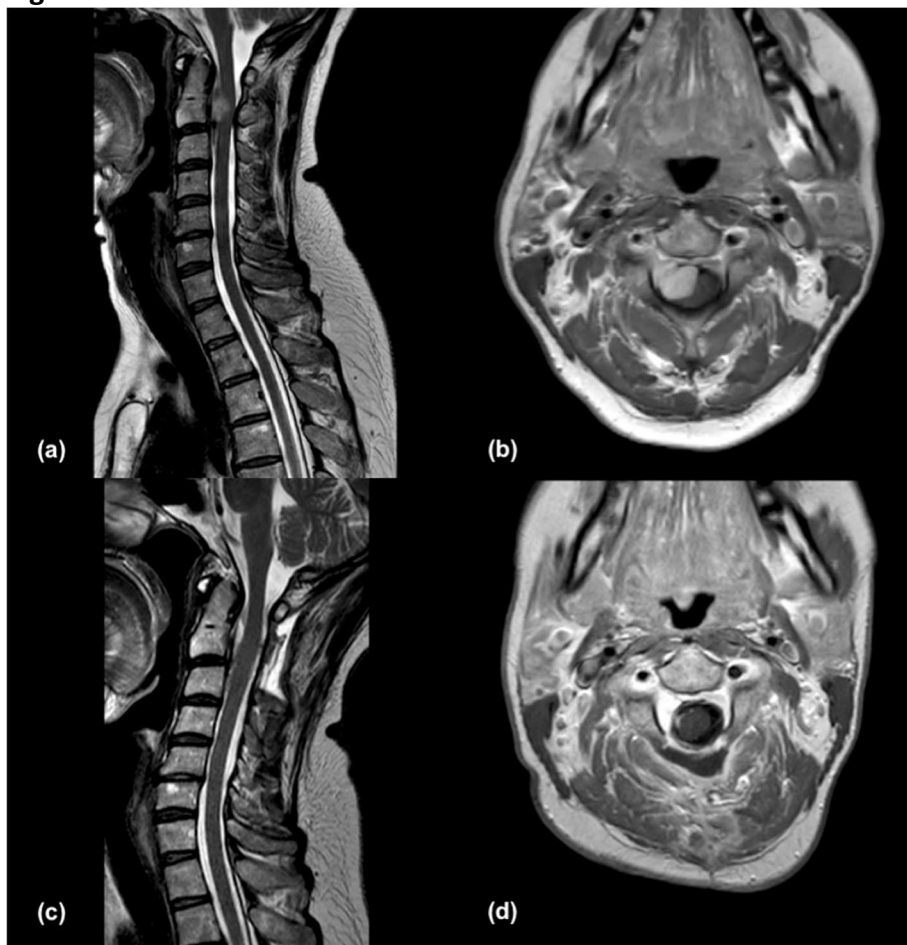


Figure 1: Sagittal T2 (a) and transverse T1+gadolinium (b) MRI imaging of the C2 meningioma at the base of the C2 vertebra compressing the spinal cord. Post operative sagittal T2 (c) and transverse T1+gadolinium (d) MRI imaging illustrating complete resection.

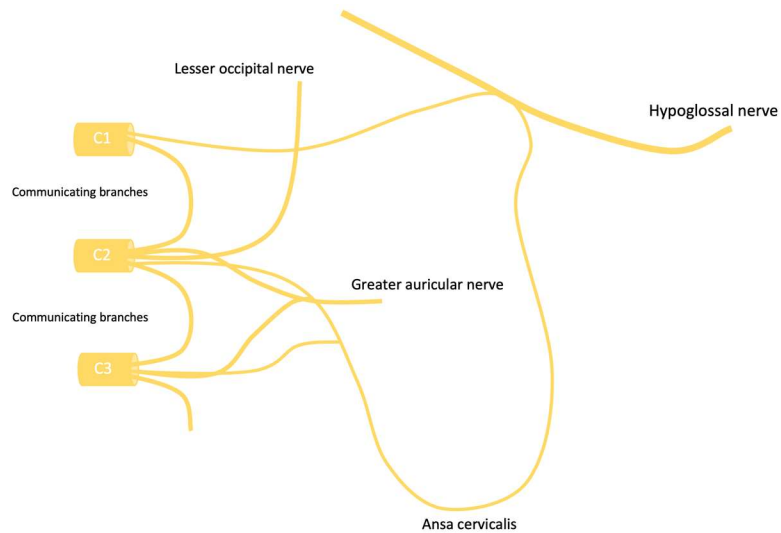


Figure 2: Schematic overview of the relation between the C2 nerve root and the hypoglossal nerve, the greater auricular nerve and the lesser occipital nerve. Based on (5).

Conflict of interest

None

Funding

Not applicable

Ethical approval

This case is a presentation of a patient treated in routine care. In consultation with the institutional review board, no ethical approval was needed.

Informed consent

The patient consented to report this case anonymously.

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