Introduction

We report a case of a 55-year-old woman with a right acoustic neuroma who developed severe macroglossia very rapidly after undergoing surgery in the lateral park bench position. Macroglossia is an infrequent but potentially lethal complication following posterior fossa surgery. The most dangerous consequence is airway obstruction, which can be life-threatening. The aetiology of postoperative macroglossia remains uncertain and has been attributed to arterial, venous and lymphatic compression, mechanical compression, or neurogenic causes. Only a few cases of postoperative macroglossia have been reported at present. This article provides new insights into aetiology and, more importantly, describes possible preventive measures and possible treatment.

Case report

Magnetic resonance imaging showed a 32-mm tumour in the right cerebellopontine angle, suspected to be a right acoustic neuroma (Figure 1), in
followed, the swelling continued to increase and the tongue started to protrude out of the oral cavity (Figure 3). Tranexamic acid, corticoids, promethazine and cetirizine were given. Allergic angioedema and hereditary angioneurotic oedema were excluded. She did not take any medication at home (no angiotensin-converting enzyme inhibitor) and was not known for allergies. Postoperative CT-scan showed no abnormalities.

CT scanning of the neck and tongue showed oedema but no arguments for pus or abscess formation at the larynx or pharynx. As the macroglossia did not improve after 2 weeks, the authors decided to place a tracheostoma, after which the tongue congestion declined spectacularly within 24 hours. Twenty-six days postoperatively, the patient left the intensive care unit. She was discharged in a good clinical condition 48 days after surgery. She had normal facial function (House-Brackmann grade 1) and the function of the tongue had recovered substantially. Oral feeding was adequate for liquid and semisolid food, but...
Since endotracheal intubation was atraumatic and angiotensin-converting enzyme inhibitors were not used, angio-oedema and local trauma were excluded. The poor response to corticoid administration made an immunological mechanism unlikely.

Venous drainage of the tongue takes place through the lingual and deep lingual veins, which ultimately end in the internal jugular vein. The venous drainage of the head and neck is via the internal jugular veins, external jugular veins and vertebral veins. Lymphatic drainage of the tongue is via the submucosal plexus, draining into the submental, submandibular and deep cervical nodes. It has been suggested that postoperative macroglossia can be caused by venous and lymphatic congestion, which is closely related to the position and the duration of the surgical procedure. This congestion can occur due to prolonged flexion of the head against the chest. The venous and lymphatic compression can also lead to thrombosis of the lingual, pharyngeal and even internal jugular veins, which can in itself cause and maintain macroglossia. In this case the patient was positioned in the lateral park bench position, in a semi to ¾ prone lateral position, with the right shoulder rotated anteriorly and flexion and rotation of the neck, creating a face-down angle. This position, with significant flexion of the neck and the right shoulder rotated anteriorly, apparently exacerbated lymphatic and venous congestion.

Local mechanical compression can also interfere with the venous and lymphatic drainage of the tongue. Mechanical compression can be caused by both the oral airway and the endotracheal tube as a result of pressure from the cannula or tube in the mouth and on the base of the tongue. In order to prevent pressure necrosis by the endotracheal tube, gauzes were placed as a bite block in front of the teeth. In the past, the authors have seen unilateral tongue swelling in patients after posterior fossa surgery. At that time, this was misinterpreted as the tongue being squeezed between the oral tube and teeth. Due to this misinterpretation more oral packings were used and this was probably restricted for solid food. Intelligibility was normal with minor restrictions in some specific sound productions (/t/1, for example). Tongue strength measured using the IOP (Iowa Oral Performance Instrument) showed decreased values for anterior and posterior maximum isometric power (Figure 4).

Tongue strength values improved over the next 28 weeks to normal level. Pathological examination of the tumour confirmed the diagnosis of acoustic neuroma.

Discussion

Macroglossia is a rare complication following posterior fossa procedures, with only a few cases reported at present. In this case, macroglossia developed immediately after endotracheal extubation, demonstrating that it may occur very rapidly after surgery. However, delayed onset up to 36 hours postoperatively has also been reported. Anaesthesiologists and surgeons involved in posterior fossa surgery should be aware of this rare but significant complication, and should be prepared to manage and monitor difficult airways.

The exact etiology of macroglossia remains unclear but seems to be multifactorial. Macroglossia has been attributed to venous and lymphatic congestion caused by the surgical position (the sitting position, extreme neck flexion), venous and lymphatic congestion caused by local mechanical compression (by teeth, oropharyngeal airway, endotracheal tube, throat pack), lingual arterial compression, angio-oedema due to immunological mechanisms or angiotensin-converting enzyme inhibitors, or to neurogenic causes.
an additional factor provoking this event. No pharyngeal packs were used since blood and secretion drainage into these gauze packs could worsen the compression on the drainage of the tongue.

The lingual artery arises from the external carotid artery and its branches run at the base of the tongue deep to the lingual veins. The endotracheal tube and packed gauze can also cause lingual arterial compression when the anterior-posterior dimension of the oropharynx is reduced by flexion of the neck. This can lead to the development of regional ischemia.

The rapid onset of macroglossia after endotracheal extubation could be related to compression ischemia of the tongue, with reperfusion hyperaemia and capillary leakage as important mechanisms.

Since macroglossia has occurred almost only after posterior fossa surgery, Moore et al. have suggested that macroglossia is a result of neurogenic mechanisms similar to neurogenic lung oedema. The pathogenesis of this neurogenic mechanism is not completely understood but it is associated with abnormal brainstem discharges provoked by surgical manipulation or tumour infiltration. Sympathic activity is said to be the cause since the brainstem is closely involved in the autonomous innervation of the tongue, the pharynx and the lowest one third of the face.

This explanation seems unlikely in our patient since the patient remained haemodynamically stable throughout the procedure and there was almost no surgical manipulation of the brainstem. Furthermore, there was no oedema or trauma of the brain stem on the postoperative CT scan.

In summary, the macroglossia in this case resulted from lymphatic rather than venous drainage blockage caused by a combination of head positioning, extreme neck flexion and the pressure of the endotracheal tube on the tongue and the floor of the mouth. Oral packing and reperfusion hyperaemia were contributory factors. We therefore advise avoiding factors that may precipitate macroglossia. Anaesthesiologists and surgeons should consider not placing the patient’s chin firmly against the chest and caution should be taken with the placement of a bite block during prolonged surgery. Oral packing should not be used. If there is unilateral swelling of the tongue, this should be considered a ‘near-miss’ case, and the position and installation of future patients should be reconsidered. If macroglossia occurs, the removal of the endotracheal tube and placement of a tracheostoma is a solution. After the placement of a tracheostoma and the removal of all pressure on the tongue and floor of the mouth, the congestion declined rapidly. In this case, the patient was positioned in the lateral park bench position, in a semi to ¾ prone lateral position, with the right shoulder rotated anteriorly and the neck in significant neck flexion in order to create space to access the posterior fossa. The authors have now reconsidered the positioning. Patients remain in a supine position, with the shoulder being abducted to 45°, which makes the extreme flexion of the cervical spine unnecessary.

**Conclusion**

Macroglossia as a postoperative complication should be kept in mind in posterior fossa surgery since it can be life-threatening. We assume that the lymphatic (and venous) drainage of the tongue was impeded due to prolonged compression caused by a combination of positioning with marked neck flexion, pressure of the endotracheal tube on the tongue and floor of the mouth and oral packing. We therefore agree with the multifactorial aetiology of macroglossia suggested in previous reports. It seems prudent to prevent factors that may precipitate macroglossia: oral packing should not be used and patient positioning should be well thought out. Unilateral swelling of the tongue should be considered a ‘near-miss’, and the position and installation of subsequent patients should then be reconsidered. If there is macroglossia, the solution is to remove the endotracheal tube.

**References**

Macroglossia after posterior fossa surgery


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