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could have proceeded as planned. Therefore, our experience suggests that preliminary tracheostomy is still the choice method for securing an airway in patients undergoing the repair of complex LeFort and skull fractures during which the face is neither exposed nor accessible to the anesthetist.

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Macroglossia: Compartment Syndrome of the Tongue?

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MACROGLOSSIA is an infrequent but potentially lethal postoperative complication following intracranial neurosurgical procedures in the posterior fossa.¹⁻¹¹ The incidence of this complication has been estimated to be 1%⁹; however, the complication may be underreported, and its actual incidence is unknown. The etiology of macro-

glossia is uncertain, and has been attributed to arterial compression, venous compression, mechanical compression, or to neurogenic origin.^{6,9} Recently, Kuhnert *et al.* described a case of massive postoperative macroglossia following a C1-2 laminectomy using the sitting position in a patient with Crouzon syndrome. Although the etiology remained unclear, the case revived interest in this complication.¹⁰

This report documents our experience with a patient who experienced this complication during her first procedure for resection of an acoustic neuroma, and who returned 9 yr later for resection of the recurrent tumor. In addition, another case of a "near miss" is described. These two cases lend new insight to the etiology of this complication, and suggest that venous obstruction leading to arterial insufficiency and resulting in reperfusion injury is the primary etiology in many of these cases.

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Case 1

A 43-yr-old overweight (170-cm, 100-kg) woman presented for left suboccipital craniectomy for an acoustic neuroma (2 x 2 cm). The patient was placed in the park-bench position (semiprone lateral with the left side up and the head flexed). A soft bite block (rolled-up gauze) was inserted after induction of anesthesia. Surgery and anesthesia were uneventful and lasted for 10 h. At the end of the procedure the patient awoke in the operating room, and the trachea was extubated shortly afterward. The tongue was noted to be large but not swollen, and the airway was patent. Thirty minutes after arrival in the recovery room the patient reported the inability to breathe, was noted to be stridulous. The tongue, lower face, and neck were swollen. Direct laryngoscopy revealed a massive swollen tongue and a diffuse edema in the base of the tongue and in the supraglottic structures, and the larynx could not be observed. After multiple attempts, the trachea was reintubated. The tongue continued to swell during the next 24 h, and eventually protruded well out of the oral cavity. To prevent desiccation, tongue was wrapped with wet gauze hourly. The submandibular area and the neck were edematous. Multiple allergy testing and investigations, including C1 esterase-inhibitor levels to rule out hereditary angioneurotic edema, were conducted with negative results. On day 9, tracheostomy and exploration of the neck were performed. The only finding was a small area of necrosis in the base of the tongue. The swelling began to subside on day 12. By day 23, the tongue had returned to near-normal size and the patient was discharged.

Nine years later the patient returned for surgery on the recurrent left acoustic neuroma. The mouth opening was limited from mild contracture, and there were dental impressions on the right border of the tongue, which appeared normal. We proceeded with nasal tracheal intubation using sedation, and placed the patient in the sitting position for the surgical procedure. In addition to a multiorifice central venous catheter, a retrograde catheter was inserted into the right jugular bulb. Simultaneous monitoring of central venous pressure and jugular venous pressure (JVP) allowed for the diagnosis of venous obstruction (should it occur); central venous pressure would remain unchanged whereas JVP would increase, thereby creating a gradient. Supine extreme flexion of the neck with the head turned to the right created a gradient of 8 mmHg. The patient was placed in the sitting position with head turned to the left to facilitate surgical exposure. A soft bite block was placed. The transducer for JVP was placed at the level of the jugular bulb (at the level of the mastoid process). Comparison with central venous pressure indicated that there was no venous obstruction, and the JVP remained negative throughout the procedure. Surgery lasted 9.5 h. No swelling was observed in the tongue or neck at the end of the procedure. After 30 min of observation the nasotracheal tube was removed. The postoperative course was uneventful.

Case 2

A 50-yr-old woman underwent a suboccipital craniectomy for the resection of a posterior fossa arteriovenous malformation. After the uneventful induction of anesthesia, the patient was placed in the sitting position. Approximately 30 s after placement of the head in the Mayfield head-holder and flexion of the neck, a blue discoloration developed above the neck. The lips were blue and the tongue began to protrude between the teeth. The pulse oximetry remained at 100%. The flexion of the neck was quickly reduced and the head was brought

back toward a more-neutral position by approximately 10°. The patient's color returned to normal almost instantaneously. The surgical procedure and the patient's recovery were otherwise uneventful.

Discussion

As recently suggested by Kuhnert *et al.*,⁶ the etiology of macroglossia may be multifactorial. Factors that have been implicated can be classified as (1) local mechanical compression (by teeth, oropharyngeal airway, endotracheal tube, throat pack) interfering with venous or lymphatic drainage of the tongue; (2) regional venous obstruction (excessive neck flexion, head-down position); and (3) various combinations of these two factors. Moore *et al.*⁹ and Ishiyama *et al.*¹¹ observed that the complication occurred only with posterior fossa surgery, and proposed that macroglossia may be neurogenic in origin; however, neither report provided evidence to support this hypothesis.

A summary of the neurosurgical cases reported in the literature, including the present case, is shown in table 1. Of the 16 cases of macroglossia reported, all but one involved posterior fossa procedures. The most plausible explanation may be that the surgical approach to posterior fossa necessitates positioning that may lead to venous obstruction. Irrespective of the position used, surgical access to the posterior fossa frequently necessitates flexion and rotation of the neck that may impinge on the venous circulation, an obstruction that cannot always be recognized by external inspection. Although the original cases reported involved the sitting position,¹ subsequent cases have been reported that use other positions (table 1).

Analysis of the onset of macroglossia in the 16 cases suggests that there are two patterns. We hypothesize that, in the milder form, there is local venous or lymphatic congestion caused either by local compression or by venous obstruction (interfering with venous drainage *via* the lingual veins), and leads to an isolated swollen tongue. In this situation, the onset of macroglossia tends to be immediate. Airway obstruction is unlikely to occur because the swelling can be immediately recognized and extubation of the trachea can be postponed. In the more severe form, there is regional venous obstruction with or without local compression.

The lingual arteries run at the base of the tongue, deep to the lingual veins that drain into the ipsilateral internal jugular vein or into the facial vein. In susceptible patients the initial venous congestion may lead to the development of regional ischemia by compressing these arteries. This is followed by reperfusion injury. The lymphatics follow the vessels and may play a contributory

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Table 1. Summary of Case Reports of Macroglossia

Authors	Year Reported	Age (yr)	Position	Duration of Procedure	Oral Airway	Areas of Swelling	Airway Compromise*	Time Delay of Onset	Time to Resolution
Lam and Vavilala	1999	43	Park bench	10 h	No†	Tongue/face/neck	Severe	30 min	23 days
Drummond ¹⁰	1999	44	Prone	9 h	No	Tongue/face	No	2 h	4 days
Kuhnert ⁶	1999	17	Sitting	7.75 h	No‡	Tongue	Severe	30 min	15 days
Pivalizza ⁷	1998	31	Prone	4 h	No	Tongue/soft palate	Severe	0 min	9 days
Figueredo-Gaspari ⁸	1997	17	Sitting	6.5 h	No†	Tongue	Severe	2 h	14 days
Ishiyama ¹¹	1990	46	Prone	7.8 h	?	Tongue/face/neck	Severe	2 h	14 days
Moore ⁹	1988	27	Park bench	14 h	No§	Tongue	Severe	30 min	14 days
	1988	32	Park bench	12 h	No§	Tongue	Severe	8 h	14 days
	1988	54	Park bench	8 h	No§	Tongue/face	Mild	36 h	"Few days"
	1988	24	Park bench	12 h	No§	Tongue	Severe, not extubated	0 min	13 days
Teeple ³	1986	56	Supine	8 h	No	Tongue—unilateral	Mild	0 min	10 days
Mayhew ⁴	1986	1.5	Sitting	5 h	No	Tongue	Severe	20 min	12 weeks
Tattersall ⁵	1984	30	Sitting	12 h	No	Tongue/face/lips	Severe	2 h	17 days, died
Ellis ²	1975	21	Sitting	14 h	Yes	Tongue/face/neck	Severe	Min?	21 days
McAllister ¹	1974	2	Sitting	6 h	Yes	Tongue	Severe, not extubated	0 min	Died
	1974	45	Sitting	10 h	Yes	Tongue	Severe, not extubated	0 min	1 day

* Severe: required immediate intervention, either reintubation or tracheostomy.

† Soft bite block used.

‡ TEE probe used.

§ Throat pack used.

|| Stridor only, patient not reintubated.

role. The venous obstruction need not be complete and cerebral swelling may or may not be apparent. Because the reperfusion injury does not occur until the venous congestion is relieved, the onset of macroglossia is often delayed by approximately 30 min or more. The injury tends to be severe, and areas other than the tongue are often involved, as in our case. Of the 16 cases reviewed and tabulated in table 1, only 30% (5 of 19 cases) are of immediate onset, and the majority (70%) are delayed.

In the first case, the patient's second procedure presented us with a dilemma. Using a jugular bulb catheter, we were able to monitor the JVP to ensure good venous drainage. Although a direct cause and effect cannot be established, the presence of low JVPs and the uneventful postoperative course suggest that venous congestion was an important etiologic factor in the events following the first procedure.

Our second case confirms that venous obstruction from excessive flexion of the neck can occur when the patient is placed in the sitting position. Here, the venous obstruction was nearly complete and immediate, and allowed the diagnosis to be made and the complication of macroglossia to presumably be prevented.

In a letter to the editor, Drummond¹⁰ reiterated the importance of using a soft bite block instead of an oropharyngeal

airway, which was suggested by McCallister *et al.*¹ Although we agree with this recommendation, we also note that it did not prevent the occurrence of macroglossia in our patient, nor did it prevent the complication in the series reported by Moore *et al.*⁹ (table 1). The soft bite block might prevent local compression of the tongue by the teeth, but cannot prevent regional venous obstruction from neck flexion. Moreover, many patients developed this complication without the use of an oropharyngeal airway (table 1). Because flexion of the neck can result in the whole spectrum of obstruction, from the absence of obstruction to complete venous obstruction, mild to moderate swelling of the tongue may occur without causing significant airway impairment. These cases may go unnoticed, let alone reported.

In summary, review of the literature and our case experiences suggest that macroglossia following posterior fossa procedures is not an infrequent complication, and is not unique to the use of the sitting position. Its etiology may be related to venous and arterial obstruction leading to reperfusion injury. The risk factors include neck flexion, obesity, local compression, and long duration. Because the onset of macroglossia may be delayed, the risk of complete airway obstruction is al-

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ways present, and anesthesiologists need to be aware of this potential life-threatening complication.

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Unusual Presentation and Treatment of Postlumbar Puncture Headache in an 11-yr-old Boy

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SPINAL anesthesia is commonly performed in the adult surgical population and less commonly in children. Al-

though complications of lumbar puncture and spinal anesthesia have been extensively reported in the adult medical literature, complications in the treatment of pediatric patients have been less frequently recognized and described.

Postlumbar puncture headache has been reported in children.¹⁻⁴ The use of an epidural blood patch to treat pediatric patients with postlumbar puncture headache^{2,5,6} and subarachnoid fistula^{7,8} has previously been described. We report an unusual presentation and the successful management of postlumbar puncture symptoms of 10-month duration in an 11-yr-old boy with cerebral palsy.

Case Report

An 11-yr-old boy with extrapyramidal cerebral palsy was admitted to our hospital for a lower gastrointestinal hemorrhage that resolved with medical therapy. During the evaluation, the patient's mother described his longstanding difficulties with postural headache, marked postural dizziness, and intermittent bilateral lower extremity shooting pains. The etiology had not been clarified during workups by pediatricians, neurologists, and neurosurgeons.

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