

Macroglossia Following Craniotomy in Semi-Sitting Position: Case Report and Review of Literature

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Abstract

Background: Macroglossia is an infrequent but potentially lethal postoperative complication following intracranial neurosurgical procedures.

Case presentation: 37-year-old female patient underwent craniotomy for sub thalamic brain tumor in sitting position lasted 10 hours, following extubation developed macroglossia with difficult intubation ended with surgical tracheostomy, complicated with sepsis and died 16 days post operatively.

Conclusion: Our case report and review of the literature, we suggest that macroglossia risk factors include neck flexion (position), obesity, local compression, and long surgical duration. The risk of complete airway obstruction is always present, and anesthesiologists need to be aware of this potential life-threatening complication because the onset of macroglossia may be delayed.

Keywords: macroglossia, craniotomy, airway intubation.

ABBREVIATIONS: Mg-milligram; Kg-kilogram; Mm-millimeter; µg-microgram; MAC-minimum alveolar concentration; % - percentage; PRBC- packed red blood cell; PaCO₂- carbon dioxide alveolar partial pressure; O₂ Sat- oxygen saturation; ml- milliliter; MmHg- millimeter mercury; ICU- intensive care unit; Bpm- breathe per minute.

BACKGROUND

Macroglossia is an uncommon and lethal postoperative complication following neurosurgical procedures mainly in the posterior fossa surgery [1-10]. The estimated incidence of this complication is around 1% [9]. However, the complication is still underreported. The etiology of macroglossia is still unclear, and has been attributed to compression (arterial, venous, or mechanical), and to neurogenic origin [6,9].

CASE PRESENTATION

A 37-year-old single female patient, known case of suprasellar brain tumor (low-grade glioma with pilocytic features), she is not known to have any allergies. She

underwent left parietal craniotomy & partial excision of the tumor in 2000 followed by chemotherapy and radiotherapy.

In 2014 she started to complain of blurred vision, her condition was diagnosed as metastatic sub-thalamic brain tumor and planned for craniotomy, biopsy and debulking of the tumor using the navigation system. The procedure lasted for 10 hours and in semi-sitting position with her neck flexed and a throat pack was inserted. The patient was invasively monitored intraoperatively with left radial arterial line and right internal jugular central line. Induction of anesthesia carried with propofol (2 mg/kg) and fentanyl (1.5µg/kg) and maintained inhalational isoflurane (1 MAC); along with muscle relaxation injecting cisatracurium (0.15 mg/kg) intubation with an endotracheal tube (internal diameter 7.5mm) and the patient was attached to continuous infusion. She was given intra-operatively 3 liters of crystalloids and one unit of PRBC transfused with adequate urine output. Consequently, her vital signs were stable all through the procedure, mean blood pressure around 65 mmHg, PaCO₂ 33 mmHg and oxygen saturation 99%.

At the end of surgery, she was given reverse of muscle relaxant, the throat pack was removed, spontaneously breathing of tidal volume 800-850ml at respiratory rate 1012bpm, stable vital signs, in supine position, extubation was done alongside, finally, she opened her eyes and moved her head. While re-do the slipped head dressing by neurosurgeon we noticed the tidal volume through face mask was decreased to about 400-450ml, spontaneously, with O₂ saturation 98-99%. Furthermore, we noticed a rapid increase in her tongue size with decrease in her tidal volume along with decrease in her O₂ Sat early 90s% and increase in the manual mask ventilation resistance. Oropharynx was checked by laryngoscope for any foreign body. Mask ventilation became harder, the patient started to have stridor & O₂ sat dropped to 80s%. We called surgical help while keeping ventilation by face mask for urgent tracheostomy.

After tracheostomy, she was transferred to the ICU and kept on mechanical ventilation and sedated (with propofol and remifentanyl). She was covered by broad spectrum antibiotics. Enteral feeding through nasogastric tube and chest physiotherapy was done regularly.

Two- days later (Figure 1), she started to have more swollen tongue. Maxillofacial team was consulted, and they applied a mouth gag. Five -days later, she started to have increase in WBC count and septic work-up was done. It revealed gram negative bacilli. Nine- days later, her creatinine level was back to normal and her tongue size back to get increase in size. Sixteen -days later, she developed asystole. So cardiopulmonary resuscitation was started for 30 minutes, unfortunately we lost our patient.



Figure 1: Patient appears with macroglossia and tracheostomy connected to ventilator tube.

DISCUSSION

Macroglossia is an uncommon and lethal postoperative complication following neurosurgical procedures mainly in the posterior fossa surgery [1-10]. The estimated incidence of this complication is around 1% [9]. However, the complication is still underreported. The etiology of macroglossia is still unclear, and has been attributed to compression (arterial, venous, or mechanical), and to neurogenic cause [6,9].

The etiology of macroglossia is suggested to be multifactorial. These factors can be classified as: mechanical compression (by teeth, oropharyngeal airway, endotracheal tube, throat pack) obstructing the venous or lymphatic drainage of the tongue; another factor is positional obstruction (due to excessive neck flexion, head position); or combinations of these two factors.

Our review of similar neurosurgical cases reported in literature as in Table 1. 36 cases of macroglossia were reported, many were in posterior fossa intracranial procedures. Our explanation may attribute to the surgical approach as positioning may lead to venous obstruction. Away from positioning, posterior fossa surgical access requires flexion and rotation of the neck altering the venous circulation, this could not be externally inspected. Although, many reported cases in literature were in sitting position, 1 also other case were in other positions (Table 1).

Our analysis of macroglossia onset in 36 reviewed cases (Table 1), suggest: severe form where venous obstruction (with or without local compression), tends to be more rapid and dangerous. The milder form, there is local lymphatic congestion (caused either by local compression or by venous obstruction) leads to swollen tongue. Airway obstruction is unlikely to happen because the swelling can be immediately recognized, and extubating of the trachea can be delayed.

CONCLUSION

In summary, review of the literature and as in our case, we suggest that macroglossia risk factors include neck flexion (position), obesity, local compression, and long surgical duration. The risk of complete airway obstruction is always present, and anesthesiologists need to be aware of this potential life-threatening complication because the onset of macroglossia may be delayed.

Table 1

Authors	Year reported	Age (yrs)	position	Duration of procedure	Oral air-way	Areas of swelling	Airway compromise*	Time delay of onset	Time to resolution
G.Vermeersch.T.Menovsky 31	2014	55 F	Lateral Park Bench	6.5 hours	Yes	Tongue	Severe	Immediately	28 weeks
Ifeanyi Iwuchukwu 29	2014	45 F	Prone	Several hours	Yes	Tongue	Mild	1 day	2 weeks
Ifeanyi Iwuchukwu 29	2014	79 F	Supine	Hours	Yes	Tongue	Mild	3 days	1 week
Monish S Raut 27	2014	65 F	Supine	No surgery	Yes	Tongue	Severe	Minutes	4 days
MC Rajesh,EK Ramads 23	2013	1 F	Supine	220 min	Yes	Tongue	Mild	30 min	3 days
H Kafali,N Necat 30	2013	27 F	Supine	1 hour	No	Tongue and facial	Severe	Few Minutes	7 days
Pradeep kumar sharma 28	2012	24 F	Supine	2.5 hours	Yes	Tongue	Mild	10 min	10 days
Nimjee et al. 26	2012	34 F	Park bench	Several hours	Yes	Tongue	Mild	Minutes	1 week
El Hassani Y, Narata AP, Pereira VM, Schaller C 18	2012	53 F	Prone	Unknown	No	Tongue	Mild	24 hours	3 weeks
Tumul Chowdhury, Nidhi Gupta, and Girja Prasad Rath 19	2012	7 M	Sitting	unknown	No	Tongue	Severe	immediately	12 days
Szu-Kai Hsu and Cheng-Ta Hsieh 14	2012	43 F	Park bench	8 hours	No	Left neck and face	Severe	2 hours	7 days
Mei-Ching Huang, Yung-Cheng Wang 15	2012	42 F	Left lateral	6.5 hours	No	Left neck and face	Severe	0 min	16 days
S.Toyama, K.Hoya 17	2012	32 M	Park bench	8.1 hours	No mouth gauzes	Tongue and face bilateral	Severe	Few minutes	33 days
Janae L.Maher,Raman C.24	2011	5 F	Supine	No surgery (traumatic)		Tongue	Mild	30 minutes	1 week
Shino Junghaenel, Titus, Robert Mischkowski 20,0	2011	1.7 F	Supine	3.5 hours	Yes	Tongue and hypo-pharyngeal	Severe	30 min	9 days
Shimizu s, K Sato, I Mabuchi, S Utsuki, H Oka, S Kan 21	2009	56 F	Left Park bench	10 hours	No	Face and neck	Severe	2 hours	10 days
Kotil, Kadir MD*; Yavasca, Pinar MD†; Bilge, Turgay 22	2006	44 M	Sitting	?	?	?	Severe	?	Partial Glossectomy
Arthur M. Lam, Monic S. avilala13	2000	50 F	Sitting	?	No	Tongue and lips	Severe	While flexion	1 day
V. Bhadri Narayan, 16	1999	40	Lateral	8 hours	Yes	Unilateral left neck and face	Severe	2 hours	21 days
Lam and Vavilal 13	1999	43	Park bench	10 hours	No +	Tongue, neck and face	Severe	30 min	23 days
Drummond 10	1999	44	Prone	9 hours	No	Tongue and face	No	2 h	4 days
Kuhnert 6	1999	17	Sitting	7.75 hours	No ++	Tongue	Severe	30 min	15 days
Pivalizza 7	1998	31	Prone	4 hours	No	Tongue and soft palate	Severe	0 min	9 days
Figeredo-Gaspari 8	1997	17	Sitting	6.5 hours	No +	Tongue	Severe	2 hours	14 days
Ishiyama 11	1990	46	Prone	7.8 hours	?	Tongue, face and neck	Severe	2 hours	14 days
Moore 9	1988	27	Park bench	14 hours	Throat pack	Tongue	Severe	30 min	14 days
Moore 9	1988	32	Park bench	12 hours	Throat pack	Tongue	Severe	8 hours	14 days
Moore 9	1988	54	Park bench	8 hours	Throat pack	Tongue and face	Mild	36 hours	Few days
Moore 9	1988	24	Park bench	12 hours	Throat pack	Tongue	Severe, not extubated	0 min	13 days
Teeple 3	1986	56 F	Supine	8 hours	No	Tongue	Mild "stridor"	0 min	10 days
Mayhew 4	1986	1.5 M	Sitting	5 hours	No	Tongue	Severe	20 min	12 weeks
JC Denny 25	1985	1.5 M	Supine	8 hours	No	Tongue	Severe	Few minutes	3 months
Tattersall 5	1984	30 F	Sitting	12 hours	No	Tongue, face and lips	Severe	2 hours	17 days died
Ellis 2	1975	21 M	Sitting	14 hours	Yes	Tongue, face and neck	Severe	Min?	21 days
McAllister 1	1974	2 F	Sitting	6 hours	Yes	Tongue	Severe, not extubated	0 min	Died
McAllister 1	1974	45 F	Sitting	10 hours	Yes	Tongue	Severe, not extubated	0 min	1 day

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

+soft bite block used.

++TEE probe used.

? Unknown.

DECLARATIONS

I declare that I have no competing interests

AVAILABILITY OF DATA AND MATERIALS

Not Applicable.

ETHICAL APPROVAL

Not Required.

CONSENT FOR PUBLICATION

Written informed consent was obtained from the kin of patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

COMPETING INTERESTS

None.

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AUTHOR CONTRIBUTION

A. AJ has written the case report and he is the doctor in charge of the patient.

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REFERENCES

1. Kuhnert SM, Faust RJ, Berge KH, and Peipgras DG. Postoperative macroglossia: Report of a case with rapid resolution after extubation of the trachea. *Anesth Analg.* 1999;88(1):220-223
2. Narayan VB, Rao GSU. Unilateral facial and neck swelling after infratentorial surgery in the lateral position. *Anesth Analg.* 1999;89(5):1290-1291.
3. Szu-Kai Hsu, Cheng-Ta Hsieh, Chih-Ta Huang, Jing-Shan Huang. Delayed airway obstruction in posterior fossa craniotomy with park-bench position. *Surg Sci.* 2012;3(11):526-529
4. Mei-Ching Huang, Yung-Cheng Wang, Jing-Shan Huang, Kou-Mou Huang. A rare presentation of hemi facial swelling following neurosurgery in lateral position: A Case Report. *Clin Mol Med.* 2012;3(1):7-9
5. S. Toyama, K. Hoya, K. Matsuoka, T. Numai and M. Shimoyama. Massive macroglossia developing fast and immediately after endotracheal extubation. *Acta Anaesthesiol Scand.* 2012;56(2):256-259.
6. Yassine El H, Narata AP, Pereira VM, Schaller C. A reminder for a very rare entity: massive tongue swelling after posterior fossa surgery. *J Neurol Surg A Cent Eur Neurosurg.* 2012;73(3):171-174
7. Chowdhury T, Gupta N, Rath GP. Macroglossia in a child undergoing posterior fossa surgery in sitting position. *Saudi J Anaesth.* 2012;6(1):85-86.
8. Junghaenel S, Keller T, Mischkowski R, Hinkelbein J, Beutner D, Koerber F, et al. Massive macroglossia after palatoplasty. *Eur J Pediatr.* March 2012;171(3):433-437
9. Maher JL, Mahabir RC, Read LA. Acute macroglossia in the pediatric patient: worth a look. *Pediatr Emerg Care.* 2011;27(10):948-949.
10. Sharma PK, Bhakta P, Srinivasan S, Khan RM, Kaul N. Acute tongue enlargement secondary to pharyngeal packing after tracheal intubation-A case report. *Middle East J Anaesthesiol.* 2012;21(5):761-764