

Case report

Ischemic necrosis of the tongue following transoral neurosurgical procedure

Amine Chakroun^{1,*}, Imen Achour¹, Lotfi Chaari², Ilhem Charfeddine¹,
Bouthaina Hammami¹, Abdelmonem Ghorbel¹

¹ Department of Otorhinolaryngology, Habib Bourguiba hospital, Sfax, Tunisia

² Department of Neurosurgery, Habib Bourguiba hospital, Sfax, Tunisia

(Received 22 October 2013, accepted 15 December 2013)

Key words:
tongue diseases /
necrosis /
macroglossia /
postoperative
complications

Abstract – Introduction: Postoperative, lingual swelling and necrosis are unusual complications that can lead to life-threatening airway obstruction. We report a case of bilateral ischemic necrosis of the tongue following transoral neurosurgical procedure. **Observation:** A Davis-Boyle mouth gag was used for tumor exposition and the operation lasted 8 h. At the end of the surgery, we noticed a bluish discoloration with a mild edema of the tongue. The patient had kept intubated in a monitored setting and intravenous steroids were administered. However, the edema increased and necrosis of the tongue appeared 72 h after the surgery. The patient underwent tracheotomy and the necrotic tissue was removed. More than 15 days passed before glossal engorgement resolved. **Discussion:** The etiology of tongue necrosis is likely related to tissue ischemia, secondary to venous or arterial obstruction. Prolonged use of the Davis-Boyle mouth gag may incur greater risk of ischemic necrosis of the tongue.

Mots clés :
pathologies linguales /
nécrose /
macroglossie /
complications
postopératoires

Résumé – Nécrose ischémique de la langue après une intervention neuro-chirurgicale trans-orale.
Introduction : Après une intervention chirurgicale, l'œdème et la nécrose de la langue représentent une complication rare qui peut entraîner une obstruction des voies aériennes mettant en jeu le pronostic vital. Un cas de nécrose ischémique bilatérale de la langue après une intervention neuro-chirurgicale trans-orale est rapporté. **Observation :** Un ouvre-bouche de Davis-Boyle a été utilisé pour aborder la tumeur et l'intervention a duré 8 h. À la fin de l'intervention, la langue présentait une couleur bleuâtre et un œdème modéré. La patiente a été gardée, intubée, dans l'unité de soins intensifs et des corticoïdes ont été injectés par voie intra-veineuse. L'œdème a augmenté et une nécrose de la langue est apparue 72 h après l'intervention. La patiente a été trachéotomisée et le tissu nécrotique excisé. L'œdème lingual a régressé en une quinzaine de jours. **Discussion :** La nécrose linguale est probablement due à une ischémie tissulaire, secondaire à une obstruction veineuse ou artérielle. L'utilisation prolongée de l'ouvre-bouche de Davis-Boyle augmente probablement le risque de nécrose ischémique de la langue.

Introduction

Ischemic necrosis of the tongue is a rare condition because the tongue has a rich blood supply. Severe glossal edema was reported after otolaryngologic procedures especially following palatoplasty [1, 2]. The etiology of the tongue edema may be related to prolonged venous or arterial obstruction resulting in tissue ischemia, or direct trauma to the lingual tissues from the mouth gag. As hematomas of the tongue have not been reported following these procedures, the likely pathogenesis relates to increased permeability of vessels resulting in lymphedema [3].

This case reports a documented bilateral tongue necrosis following transoral neurosurgical procedure caused by Davis-Boyle mouth gag prolonged use. To our knowledge, this is the first reported case of bilateral tongue necrosis secondary to Davis-Boyle mouth gag use.

Case report

A 45-year-old woman with foramen magnum meningioma was brought to the operation room for excision. She was intubated via an oral approach by use of a 7 mm endotracheal tube

* Correspondence: amine_chakroun@yahoo.fr



Fig. 1. A Davis-Boyle mouth gag.
Fig. 1. Ouvre-bouche de Davis-Boyle.

without any difficulty. The patient was placed in the horizontal position with the neck hyperextended. A Davis-Boyle mouth gag (Fig. 1) was placed in atraumatic fashion by use of the large-sized grooved endotracheal tongue blade to secure the endotracheal tube and retract the tongue out the operative field. The meningioma was excised through transpalatal approach under microscopic view. The neurosurgical procedure was uneventful and lasted approximately 8 h. The mouth gag was in place throughout the entire time. At the end of the surgery, after the relapse of the Davis-Boyle mouth gag and before extubation, we noticed a bluish discoloration with a mild edema of the tongue.

This edema did not seem to resolve after 30 minutes of observation in the operating room. The patient was kept intubated and brought to the intensive care unit for close observation. Heavy doses of steroids were started. Despite this therapy, the patient's tongue continued to enlarge and started protruding out of the mouth. There was no evidence of an allergic reaction, such as a rash, generalized edema, or erythema and the patient was otherwise hemodynamically stable.

Intensive care unit records observe that on further questioning, the patient denied any allergies or autoimmune disease. She also denied use of nonsteroidal anti-inflammatory drugs, aspirin or angiotensin-converting enzyme inhibitor-I and was not aware of any personal or familial history of facial swelling or angioedema.

A computed tomographic mandible scan revealed a correctly positioned laryngeal tube and a markedly enlarged tongue with anterior protrusion out of the oral cavity (Fig. 2). The patient developed subsequently a large necrotic slough of the dorsal surface of the tongue 72 h after the surgery (Fig. 3). The patient was brought to the operating room. While sedated, a tracheotomy was performed, the tube was removed and the

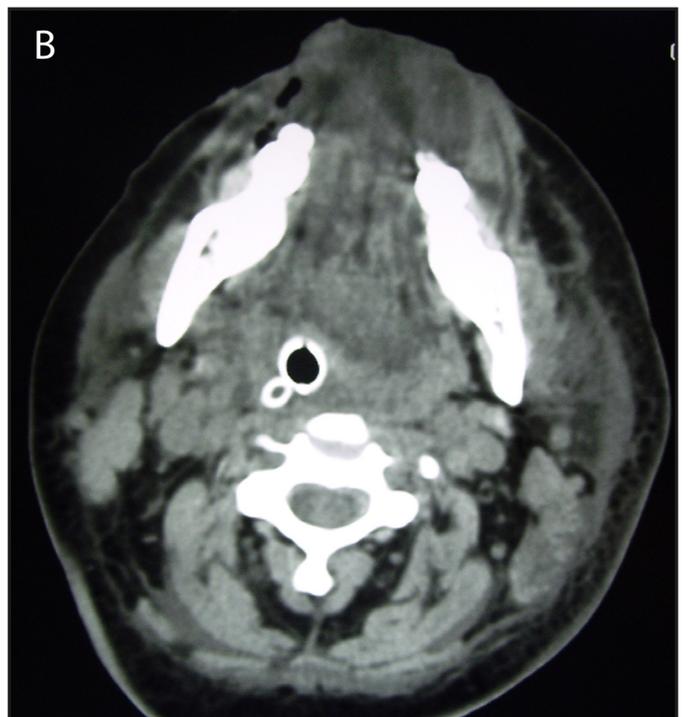
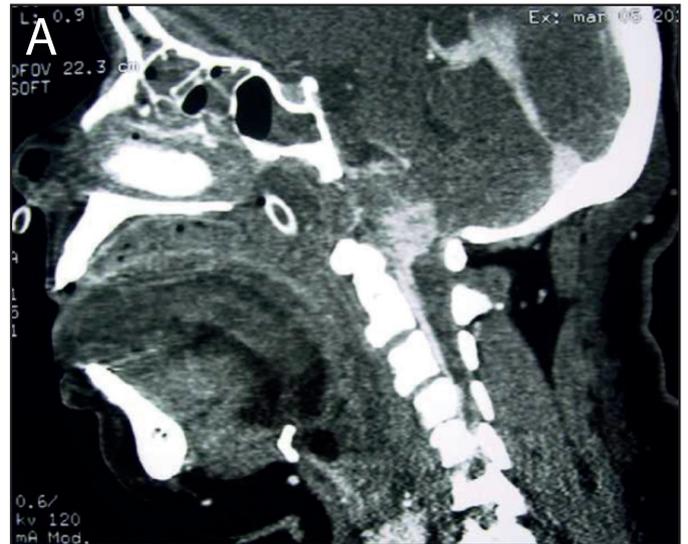


Fig. 2. Sagittal (A) and axial (B) CT image demonstrating swelling of the tongue and its protrusion out of the oral cavity. The endotracheal tube is correctly placed.

Fig. 2. CTscan (A : coupe sagittale ; B : coupe axiale) montrant l'œdème de la langue et sa protrusion hors de la cavité buccale. Le tube endo-trachéal est correctement placé.

necrotic tissue of the tongue was excised. More than 15 days passed before glossal swelling resolved completely (Fig. 4).

The histological examination of the necrotic tissue of the tongue revealed no vasculitis, or amyloidosis or calcification.



Fig. 3. 72 h after completion of neurosurgical procedure, immediately after tracheotomy. Anterior view showing severe macroglossia with necrosis of the dorsal surface of the tongue.

Fig. 3. 72 h après l'intervention chirurgicale et immédiatement après la trachéotomie : importante macroglossie avec nécrose du dos de la langue.

Discussion

Postoperative, massive, lingual swelling and necrosis are unusual complications that can lead to life-threatening airway obstruction [4]. Macroglossia and unilateral ischemic necrosis of the tongue have been reported during neurosurgical procedures [5]. The authors postulate that the tongue was severely compressed by the endotracheal tube which causes necrotic slough and/or the swelling of the tongue may have resulted from venous occlusion of the tongue base with increased tongue size and secondary compression of the tongue against the tube in a confined space. Ellis et al. suggested that an extreme flexion of the head against the chest during neurosurgical procedures may additionally compress airway, tube and tracheal rings against the base of the tongue [6]. However, in our case, the patient was placed in the horizontal position with the neck hyperextended.

Postoperative severe glossal edema can occur after palatoplasty. In literature, 5 documented cases are reported since 1950. It is hypothesized that the edema is a direct result of compression of the tongue by the Dingman mouth gag. Too much compression can cause pressure necrosis of the glossal tissue and impede lymphatic and venous outflow from the tongue, resulting in significant glossal edema [2]. In addition,



Fig. 4. 7 days after tracheotomy: the glossal edema has resolved.

Fig. 4. 7 jours après la trachéotomie : l'œdème lingual a disparu.

the classic Rose positioning of the head with the neck extended further reduces venous and lymphatic drainage [7].

The Davis-Boyle mouth gag is a device that allows for excellent visualization and access to the palate and oropharynx by retracting the tongue inferiorly under the endotracheal tube and stabilizing the anterior maxillary alveolus. Excessive retraction, in conjunction with hyperextended neck, can impair the venous and lymphatic drainage of the tongue and results in mucosal edema [1].

Theoretically, significant arterial occlusion edema would occur only if the pressure generated by the mouth gag was transferred to a major feeding artery and was above that artery's mean arterial pressure. Similarly, significant venous occlusion edema would occur only if the pressure generated by the mouth gag was transferred to a major drainage vein and that pressure was above the venous pressure.

Senders and Fung studied the pressures generated by Dingman mouth gag and McIvor mouth gag use. The mean pressure at maximum opening was 104 mmHg which is near the maximum mean arterial pressure. Suspension added an average increase of 102 mmHg. This was in addition to the pressure already generated by having the mouth gag open. They suggested that suspension of the mouth gag results in venous and arterial ischemia and that the area of ischemia would be determined by the mouth gag blade size, shape and size of the oral cavity, as well as the blade's angulations [4].

Another study suggested that procedures lasting more than 2 h have a higher incidence of postoperative lingual edema [8]. This may explain the greater incidence of this lingual edema after palatoplasty or pharyngeal flap surgery than after tonsillectomy and adenoidectomy [4].

Despite this data, postoperative severe macroglossia are very rare. In our experience, this is the first time that postoperative ischemic necrosis of the tongue occurred although we used the same surgical technique with two other patients without having this complication. We believe that ischemic necrosis of the tongue can be explained by the length of the surgical procedure in addition to differences in dentofacial structure among patients.

Prevention of glossal edema requires minimizing the compression of the tongue while allowing for visualization of the surgical field which may be difficult to achieve [2]. Some authors recommend that the tongue retractor should be released hourly for 5 to 10 minutes to allow for decompression and lymphatic drainage of the tongue. An extreme Trendelenburg position should be avoided as long as possible. An appropriately sized blade in relation to the oral cavity should be chosen. They further suggest that the tongue should be carefully examined before extubation and after removal of the mouth gag [1, 2, 4]. If glossal edema does occur, Gupta *et al.* recommend to keep the patient under close observation postoperatively for any kind of obstruction [1], while Aziz and Ziccardi suggest that the patient should be kept intubated and administered intravenous steroids until the edema resolves [2].

In our case, these measures appeared to be ineffective. The glossal edema increased rapidly despite the administration of intravenous steroids. All the dorsal surface of the tongue was necrotic. We think that the endotracheal tube kept after surgical procedure contributes to compromise venous drainage by

carrying a continuous pressure on the edematous tongue in a confined space. This hypothesis likely explains why our patient improved only after extubation and excision of the necrotic tissue.

Competing interests: none

References

1. Gupta R, Chhabra B, Mahajan RK, Nandini. Macroglossia following palatoplasty causing upper airway obstruction: case report. *J Oral Maxillofac Surg* 2001;59:940-1.
2. Aziz SR, Ziccardi VB. Severe glossal edema after primary palatoplasty. *J Oral Maxillofac Surg* 2009;67:1326-8.
3. Senders CW, Fung M. Factors influencing the palatoplasty and pharyngeal flap surgery. *Arch Otolaryngol Head Neck Surg* 1991;117:542-5.
4. Senders CW, Eisele JH. Lingual pressures induced by mouthgags. *Int J Pediatr Otorhinolaryngol* 1995;33:53-60.
5. Teeple E, Maroon J, Rueger R. Hemimacroglossia and unilateral ischemic necrosis of the tongue in a long-duration neurosurgical procedure. *Anesthesiology* 1986;64:845-6.
6. Ellis SC, Bryan-Brown CW, Hyderally H. Massive swelling of the head and neck. *Anesthesiology* 1975;42:102-3.
7. Greene AK, Kreuter M, Mulliken JB. Professor Edmund Rose: his position in surgery. *Plast Reconstr Surg* 2003;111:383-6.
8. Schettler D. Intra- and postoperative complications in surgical repair of clefts in infancy. *J Maxillofac Surg* 1973;1:40-4.