

Macroglossia secondary to prolonged prone positioning for management of COVID-19 respiratory failure

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SUMMARY

We report the case of a 59-year-old woman who developed acute macroglossia following prolonged prone positioning for management of COVID-19. We found that Biotene mixed in glycerin was effective at removing keratinised lingual plaques and better tolerated than Biotene alone. Additionally, uniform tongue compression applied via tubular elastic dressing yielded more efficacious results than uneven tongue compression via Coban.

BACKGROUND

Macroglossia or enlarged tongue¹ is a rare and potentially life-threatening² condition clinically defined as tongue protrusion beyond the teeth or alveolar ridge at rest.^{3 4} It may lead to complications such as pain, chronic tongue drying, superimposed infection, impaired speech, swallow dysfunction and even airway obstruction.⁵ Etiologies include endocrinological, metabolic or neoplastic infiltration; infectious or inflammatory conditions; trauma; and, rarely, prolonged prone positioning.⁶ There are currently no standard guidelines for the management of acute macroglossia. Here, we report the case of a 59-year-old woman who developed acute macroglossia following prolonged prone positioning for management of COVID-19.

CASE PRESENTATION

A 59-year-old woman with obesity, type 2 diabetes mellitus, obstructive sleep apnoea and history of thyroid adenocarcinoma status post thyroidectomy with surgical-induced hypothyroidism was hospitalised for COVID-19-induced

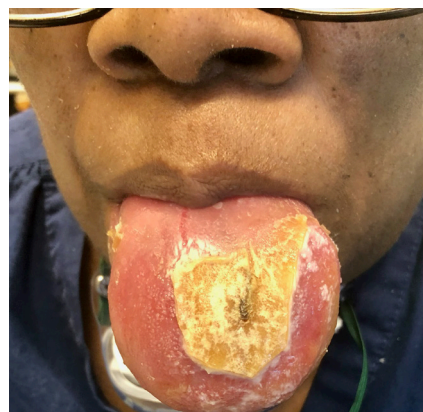


Figure 2 Twenty-four hours postdebridement with keratin reaccumulation.

respiratory failure. Acute care management included convalescent plasma, tocilizumab, intubation and mechanical ventilation for over 33 days with prone positioning for a total of 11 days (table 1). Macroglossia was noted on day 8 (table 1) of prone positioning, and a bedside tracheostomy was subsequently placed to bypass the oropharynx. Macroglossia was treated with Biotene gauze overlaid by Coban dressing and a Decadron taper. Prior to discharge from acute care, thyroid hormone assays were obtained revealing an elevated thyroid-stimulating hormone of 8.15 mIU/L in the setting of a

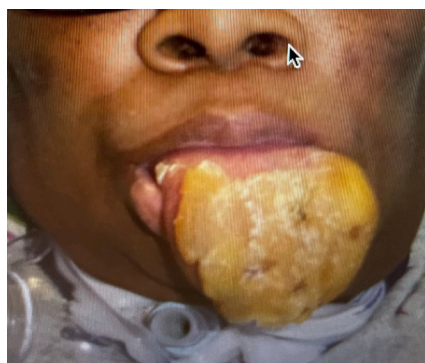


Figure 1 Admission day with macroglossia and keratinised plaque.



Figure 3 Dental bite block, superior view.



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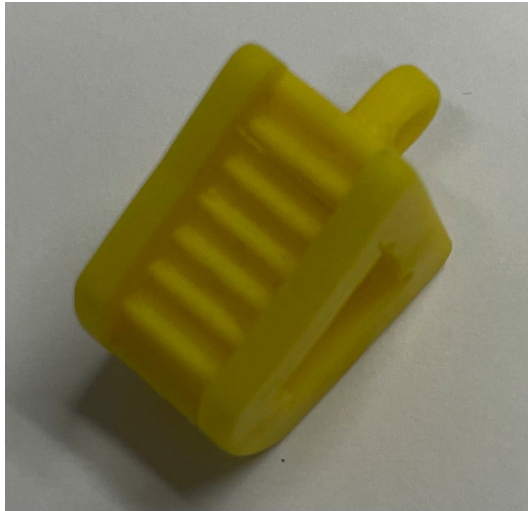


Figure 4 Dental bite block, lateral view.

normal free thyroxine level of 0.9 ng/dL. These findings suggested recovery from non-thyroidal illness in the absence of untreated hypothyroidism.

Following her 42-day acute care course, she was transferred to our facility for intensive inpatient rehabilitation. At this time, Coban dressing was discontinued due to intolerance. On arrival, she was unable to speak due to severe macroglossia, with her tongue protruding 10 cm beyond the oral cavity, 6 cm wide and 4 cm thick (figure 1). The tongue was encased in a keratinised plaque of dried-up necrotic tissue (figures 1 and 2). Recommendations from ear, nose and throat doctor were to use bite blocks (figures 3 and 4) to facilitate lymphatic and venous drainage or to have the patient undergo a partial glossectomy. The smallest bite guard (figures 3 and 4) available was introduced but subsequently removed due to intolerance for more than 5 min.

Despite debridement, keratinised plaque typically reaccumulated (figure 5). Dressings consisting of a gauze soaked in a mixture of Biotene and glycerin were placed around her tongue and covered with tubular elastic dressing for graded, even compression (figure 6). The dressings were changed twice a day. The tubular elastic dressing was suspended against gravity by two strings in order to mitigate vascular congestion by facilitating forward drainage through the normal venous and lymphatic channels (figures 6 and 7). Dressing changes with Biotene and glycerin-soaked



Figure 5 Keratin softens with use of Biotene and glycerin.

gauze overlaid by suspended tubular elastic dressing were continued twice daily, allowing her to exercise her tongue for 30–45 min prior to each dressing change. The overall goal was to keep the tongue moist and elevated while applying even compression.

OUTCOME AND FOLLOW-UP

By day 8 of her acute rehab stay, the size of her tongue was significantly reduced; keratinised tissue was completely softened and easily removed, exposing normal-appearing tongue surface (figure 8). By day 10, her tongue was clean with the exception of a small amount of debris, which was cleaned with ease. At the time of discharge, on day 14, she was able to move her tongue freely and to intermittently retract it within the oral cavity (figure 9). By day 3 postdischarge, she was able to keep her tongue inside her oral cavity and communicate normally (figure 10).

DISCUSSION

Macroglossia has been described in the literature as a rare complication of spinal and neurosurgical procedures, particularly those in the prone, park-bench and sitting positions as well as those lasting longer than 8 hours.⁵ Proposed pathophysiology includes mechanical obstruction of lymphatic, venous and arterial flow; local ischaemia and reperfusion

Table 1 Total hours prone and ventilation settings for each day of proning*

Day	1	2	3	4	5	6	7	8	9	10	11
Total hours	11.25	20.12	16	10.85	12	14	13	9.82	22.57	15	12
VT	5.14	6.25	5.27	5.49	5.42	4.83	5.3	5.36	6.87	5.33	4.83
MV	7.9	10.3	8.9	9.1	9.2	8.3	9.4	10	9.5	9.3	9.9
Rate	26	28	28	28	28	28	32	32	32	30	30
PEEP	14	20	20	28	22	18	18	18	18	18	18
PeakP	28	37	32	32	35	33	33	33	30	30	30
Pplat	26	34	30	29	32	28	30	29	29	29	27
MAP	18	26	24	24	27	22	22	22	23	22	22
FiO2	60	60	60	60	70	100	90	60	65	40	40

*Assist-control volume-control mode ventilation.

FiO₂, fraction of inspired oxygen (%); MAP, mean airway pressure (cmH₂O); MV, minute volume (L); PeakP, peak pressure (cmH₂O); PEEP, positive end-expiratory pressure (cmH₂O); Pplat, plateau pressure (cmH₂O); Rate, mechanical rate (breaths/minute); VT, tidal volume (mL/Kg).



Figure 6 Tubular dressing soaked in Biotene and glycerin.

injury.⁵ Patients with COVID-19 receiving prolonged prone-position ventilation are especially at increased risk,⁷ and there are no guidelines for managing such patients in whom standard lingual compressive therapy has failed. This case highlighted an effective method of lingual compression in patients with post-COVID-19 macroglossia.

This case highlights the heterogeneous clinical nature of macroglossia in patients following prolonged prone positioning. These patients may present with macroglossia of various sizes, with or without varying complications such as keratinised lingual plaques or infection. This heterogeneity may account for patients' varying responses to management options including corticosteroids, bite blocks (figures 3 and 4) and compressive therapy.⁸⁻¹⁰

Our patient's history of obesity and obstructive sleep apnoea may have contributed to her predisposition to severe macroglossia with prolonged prone positioning. Furthermore, she presented to our facility with a significant degree of keratinised lingual plaque formation encasing her tongue, which likely contributed to the lack of significant reduction in lingual size despite several weeks of treatment at the outside facility with Biotene-soaked gauze wrapped with a Coban compression dressing. A bite block (figures 3 and 4)



Figure 8 Ten days postadmission.

was further not able to be tolerated due to her significant degree of macroglossia.

Biotene is a moisturising oral rinse used to relieve dry mouth symptoms. Although it contains glycerin, a sweet-tasting topical humectant, Biotene has a strong taste that was not tolerated by our patient. To address this, pure glycerin was added to Biotene, making it more palatable and better tolerated by our patient than Biotene alone. Biotene mixed with pure glycerin was found to be effective in removing the keratinised lingual plaques from her tongue in as little as 3 days. Additionally, uniform tongue compression applied via tubular elastic dressing yielded more efficacious results than uneven tongue compression via Coban. We found that removal of the keratinised layer encasing her tongue allowed for more effective tongue compression. Furthermore,



Figure 7 Dressing suspended against gravity for even, graded compression.



Figure 9 Discharge day, 2 weeks postadmission.



Figure 10 3 days after discharge.

compression against gravity more effectively facilitated drainage into the normal venous and lymphatic drainage channels than compression, which was not against gravity.

Learning points

- ▶ Biotene diluted with glycerin is an effective and tolerated means of removing keratinised lingual plaques in patients who do not tolerate Biotene alone.
- ▶ Uniform tongue compression may be more effective than uneven tongue compression in macroglossia management.
- ▶ Lingual compression against gravity may more effectively facilitate lingual drainage than compression that is not against gravity.

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