Novel treatment (new drug/intervention; established drug/procedure in new situation)

Case report

Lingual compression for acute macroglossia in a COVID-19 positive patient

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SUMMARY
We describe a 40-year-old woman with severe, persistent macroglossia following prone positioning as part of treatment for COVID-19. We used the treatment method of lingual compression with satisfactory results.

BACKGROUND
Acute macroglossia is an uncommon and perplexing clinical problem. Previously cited aetiologies include trauma, medication side effects, allergic reactions, patient positioning, pressure packing of the oropharynx and oral surgery. 1 8 It has been well described in the literature and is often self-resolving with conservative treatment including intravenous steroids and a bite block. 2 4 9 Extreme cases may lead to dysphagia and airway compromise. In certain cases, physicians have resorted to partial glossectomy, a potentially morbid management strategy, associated with postoperative speech and swallowing difficulties. 10 11 Non-operative management strategies may be a desirable alternative given the potential for postoperative morbidity.

CASE PRESENTATION
The patient is a 40-year-old African American woman with a medical history of insulin dependent diabetes mellitus, pancreatic insufficiency, asthma, hypertension and ulcerative colitis. She presented to the emergency department with 1 week of fevers, shortness of breath and diarrhoea. The patient was found to be positive for COVID-19. Her respiratory function quickly worsened despite breathing treatments. She was intubated and admitted to the intensive care unit.

The patient was treated with a combination of hydroxychloroquine, methylprednisolone and tocilizumab. Despite medical management, she developed persistent, severe hypoxia and an acute respiratory distress syndrome-like picture with a partial pressure of oxygen to fraction of inspired oxygen (PaO2:FiO2) ratio <100. In order to improve oxygenation, prone positioning was used for a total of 11 days. Our institutional prone positioning protocol includes 16 hours of prone positioning, followed by 8 hours of supine positioning. Worsening lower facial oedema and macroglossia was noted as a result of patient positioning; however, the benefits of improved oxygenation were thought to outweigh any risks. At the conclusion of prone positioning, the patient’s macroglossia was severe, with the tongue nearly capable of resting on her chest. Thyroid stimulating hormone (TSH) was sent and noted to demonstrate a relatively hyperthyroid state at 0.10 uIU/mL. A free T4 was then checked reflexively and was normal at 1.22 ng/dL. Growth hormone was not checked. We attributed the patient’s macroglossia solely to prolonged prone positioning.

Otolaryngology was consulted. An additional 10-day course of methylprednisolone in addition to a bite block was recommended. However, this yielded only mild improvement. The patient’s tongue remained significantly edematous, protruding from the mouth with pressure injuries developing over the ventral surface where the tongue was in contact with teeth and the endotracheal tube. Tracheostomy was performed on hospital day 26 after approval to perform the procedure in the setting of active COVID-19 infection was obtained from intensive care unit leadership.

The patient’s macroglossia persisted despite our interventions. In an effort to avoid partial glossectomy, lingual compression was applied on hospital day 27. The wrap consisted of saline moistened gauze loosely wrapped around the tongue, followed by a compressive Coban wrap (3M, St. Paul, Minnesota, USA) applied distal to proximal (figure 1). Care was taken to not create a lingual tourniquet. Initially, the wrap was applied once daily and left in place for 12 hours at a time. Marked improvement was observed after the first wrapping and we continued to note improvement after subsequent treatments. However, lingual oedema was noted to worsen during the 12 hours periods following treatment. On treatment day 8, lingual compression was applied twice daily for a period of 8 hours, followed by 4 hours without lingual compression.

OUTCOME AND FOLLOW-UP
On treatment day 11, the patient was able to retract the tongue into her mouth without difficulty and lingual compression was discontinued (figure 2). There was no evidence of persistent injury to the tongue as a result of treatment. The patient was discharged to a long-term assisted care facility on hospital day 52.

DISCUSSION
Acute-onset macroglossia can present significant challenges and distress to the patient when unresponsive to traditional conservative measures. The most commonly cited treatment methods include intravenous steroids and bite blocks. A tracheostomy...
Acute macroglossia is a rare clinical problem that can be distressing to both patients and providers. Acute macroglossia is typically managed conservatively with steroids and bite blocks. If macroglossia is refractory or if partial glossectomy is being considered, lingual compression may be safely used with satisfactory results. A scheduled protocol with 8 hours of compression followed by 4 hours without compression is safe and effective.

**Learning points**

- Acute macroglossia is a rare clinical problem that can be distressing to both patients and providers.
- Acute macroglossia is typically managed conservatively with steroids and bite blocks. If macroglossia is refractory or if partial glossectomy is being considered, lingual compression may be safely used with satisfactory results.
- A scheduled protocol with 8 hours of compression followed by 4 hours without compression is safe and effective.

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