

ACUTE EDEMA OF THE TONGUE: A LIFE-THREATENING CONDITION

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Acute macroglossia is rare, but may cause upper airway obstruction requiring emergency intervention. The cause of the problem is often obscure. Edema of the tongue may be due to angioedema or to allergy. We present several cases of acute edema of the tongue, in 3 cases causing life-threatening airway obstruction. Among these, we present the first case of acute enlargement of the tongue due to the ingestion of artichoke.

KEY WORDS — allergy, macroglossia, respiratory obstruction.

INTRODUCTION

Macroglossia is a relatively rare condition^{1,2} and may be congenital or acquired. The rich blood supply of the tongue from the lingual artery and its branches enables edema of the tongue to cause massive swelling.

Congenital macroglossia may be due to a variety of conditions, including hemangioma, lymphatic vascular malformations, Beckwith-Wiedemann syndrome, Down syndrome, and hypothyroidism.³

Hemangioma is a benign lesion of blood vessels or vascular elements, often appearing shortly after birth, and tending to occur in the tongue and the floor of the mouth. A hemangioma of the tongue may affect a part of the tongue or the entire tongue, producing macroglossia. When many proliferating endothelial cells line small capillaries, the lesion is referred to as a capillary hemangioma, whereas in the case of large dilated blood sinuses, the term cavernous hemangioma is applied. Most hemangiomas do not require treatment and regress spontaneously during childhood. Lymphatic vascular malformations or lymphangiomas are congenital malformations of lymph tissue that result from the failure of lymph spaces to connect to the rest of the lymphatic system. Occurrence in the oral cavity is rare, and the tongue is the usual site. Tongue lesions are amenable to CO₂ laser treatment. Beckwith-Wiedemann syndrome includes macroglossia, exophthalmos, gigantism, hyperinsulinemia, and hypoglycemia. Congenital macroglossia due to these conditions is slow-growing and uncommonly causes airway obstruction.

Acquired macroglossia can be gradual or acute. Gradual acquired macroglossia can be due to tumor infiltration, amyloidosis, or myxedema. Acute en-

largement of the tongue is infrequently encountered, and the cause of the problem is often obscure. Renahan and Morton² divided acute swelling of the tongue into 4 categories: hemorrhage, infection, infarction, and edema. Edema of the tongue may be due to angioedema or to allergy to a variety of allergens, including food.⁴⁻⁶

In 0.1% to 0.2% of patients, angiotensin converting enzyme (ACE) inhibitors induce a rapid swelling in the nose, throat, mouth, glottis, larynx, lips, and/or tongue. This angioneurotic edema apparently is not dose-related and nearly always develops within the first week of therapy, usually within the first few hours after the initial dose. The mechanism is unknown, but it may involve accumulation of bradykinin, induction of tissue-specific autoantibodies, or inhibition of complement 1 esterase inactivator. Once the patient stops taking ACE inhibitors, angioneurotic edema disappears within hours.⁷ Airway obstruction and respiratory distress may lead to death, and asphyxia due to ACE inhibitor-mediated edema of the tongue has been described.^{8,9}

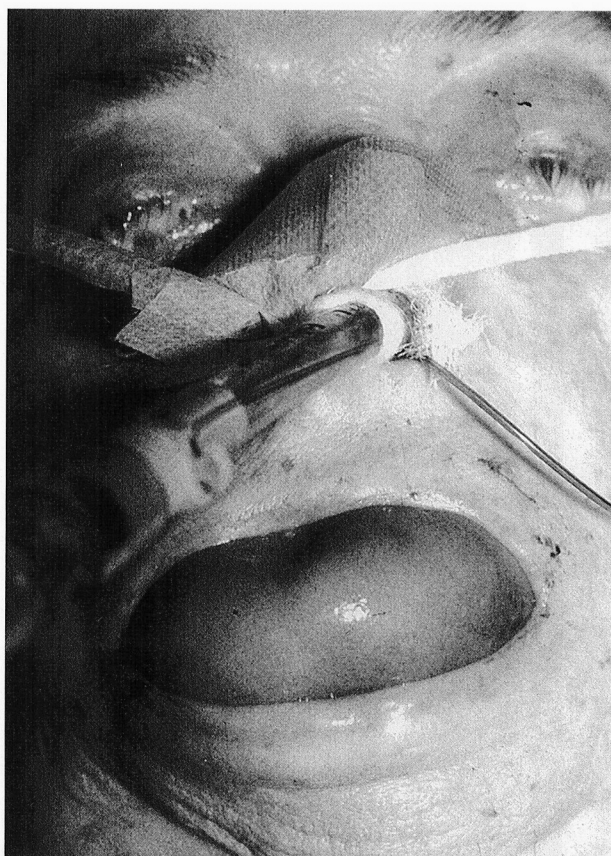
Allergy to ingestion of artichoke is rare, and has only recently been described. Patients were skin-tested, and specific IgE in serum was detected. Cases of allergic rhinitis, asthma, and urticaria have been reported, but never tongue enlargement.^{10,11}

We present several cases of acute allergic enlargement of the tongue, in 3 cases causing life-threatening airway obstruction.

CASE REPORTS

Case 1. A 91-year-old woman was brought to the emergency room from her retirement home because of severe respiratory distress caused by acute enlarge-

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(Case 1) Gross enlargement of tongue. Nasotracheal tube is in place.

ment of the tongue after a meal. A nasotracheal tube was inserted in order to secure the upper airway, and a gradual improvement was observed after systemic treatment with corticosteroids (see Figure).

Case 2. A 70-year-old woman was admitted to the emergency room in terminal condition. Her tongue was grossly enlarged and filled the entire oral cavity. Peripheral cyanosis was prominent. Tracheal intubation was not possible, and emergency cricothyroidotomy was performed. The patient survived, but brain damage was unavoidable. It was later revealed that the onset of the enlargement of the tongue was sudden, and followed the ingestion of an artichoke.

Case 3. A 65-year-old morbidly obese man was admitted to the emergency room in respiratory distress due to acute enlargement of the tongue for no apparent reason. The family later reported several similar, but less severe, episodes within the previous year. The patient was hypertensive and was treated with ACE inhibitors on a regular basis. Tracheal intubation was impossible, and therefore, the patient was taken directly to the operating room. A percutaneous tracheostomy was performed under local anesthesia. The patient was observed in the intensive care unit and gradually improved after systemic treat-

ment with corticosteroids.

Cases 4 to 10. Seven other patients suffering from acute allergic enlargement of the tongue were admitted to our hospital during the past 3 years. The ages of these patients ranged from 42 to 73 years, with an average of 56 years. In these cases, the onset of edema was gradual, and no emergency intervention was required in order to secure the upper airway. No obvious connection to the ingestion of any food was established. The patients were treated with subcutaneous administration of epinephrine and systemic administration of corticosteroids.

DISCUSSION

We have presented several cases of acute enlargement of the tongue, some obviously caused by food allergy and others of uncertain cause. Interestingly, all of the patients were more than 40 years old. Perhaps this population is at greater risk for acute macroglossia, but the reason remains unclear.

Allergy to ingestion of artichoke is rare, and has only recently been described. Cases of allergic rhinitis, asthma, and urticaria in response to artichoke have been described, but we have presented what to our knowledge appears to be the first case of life-threatening enlargement of the tongue due to artichoke allergy.

The first priority in these cases is maintenance of the airway. Three of our patients arrived with a compromised airway necessitating immediate intervention. In 1 case, an emergency cricothyroidotomy was performed, as endotracheal intubation was impossible. In another case, emergency percutaneous tracheostomy under local anesthesia was performed.

Control of swelling may be obtained by the systemic administration of corticosteroids. The most appropriate dosage seems to be 500 mg of hydrocortisone administered intravenously upon arrival to the emergency room and 100 mg of hydrocortisone subsequently given 3 times per day.

In our experience, subcutaneous administration of epinephrine in doses of 0.5 mg repeated as necessary once or twice at 15- to 20-minute intervals or until the airway is secured is also beneficial in these cases if there is no contraindication to its administration.

CONCLUSIONS

Any case of acute enlargement of the tongue should be regarded as a life-threatening condition and treated promptly. Adequate observation in an intensive care unit is necessary in all cases. Such patients should

not, by any means, be discharged from the emergency room. Maintenance of the airway is a priority. In the presence of signs of impending airway compromise, some form of artificial airway must be established. Systemic administration of corticosteroids and epi-

nephrine seems to be effective management. Investigation should be directed toward identifying the underlying cause, and subsequent treatment should be directed toward correcting that cause in order to avoid recurrence.

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