# Unilateral angioedema of tongue following ginseng containing multivitamin intake in patient with SLE: a case report

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#### ABSTRACT

Unilateral angioedema of tongue is a rare presentation of allergic reaction. If not identified and treated immediately, may lead to compromise of the airway and warrant tracheostomy or sometimes may also lead to death. In lupus patients, angioedema may be due to an acquired type of C1 inhibitor deficiency. Acquired angioedema can also be drug induced mostly seen with the use of non-steroidal anti-inflammatory drugs, contrast media, proton pump inhibitors and serotonin reuptake inhibitors. There are rare cases reported of angioedema related to Ginseng intake.

Keywords: angioedema, lupus erythematosus, panax, systemic

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#### INTRODUCTION

Angioedema is sudden swelling of the dermis, subcutaneous tissue, mucosa, and submucosa. It often presents on the face and oropharynx, often with a risk of airway compromise or may end into anaphylaxis, which is a life-threatening medical emergency.<sup>1</sup> Angioedema can be either histaminic mediated or bradykinin mediated. Histamine mediated presents with skin hives and anaphylaxis and bradykinin might present slowly with involvement of face that could progress to involve the airways.<sup>2</sup> Angioedema can also be divided into hereditary or acquired. The latter is often druginduced: associated with the use of non-steroidal anti-inflammatory drugs, contrast media, statins, proton pump inhibitors and serotonin reuptake inhibitors. In lupus patients, angioedema is the result of an acquired type of C1 inhibitor deficiency, most probably due to antibody formation directed against the C1 inhibitor molecule.<sup>1,3</sup> There are controversies about the rate of allergy in patients with lupus. Some studies have shown no difference in allergic disease among patients with lupus as compared to general population, whereas some have shown a significant increase of incidence of allergic reaction.<sup>4</sup>

Ginseng is being used in various tonics and supplements in different parts of the world5. There are cases reported about anaphylactic reaction not only to oral intakes but also from inhalation6.

#### **Case Presentation**

Forty-five year's female presented to the Emergency Room (ER) following swelling of tongue and difficulty in swallowing for 2 hours which started after taking ginseng containing antioxidant and multimineral capsule, prescribed a day before when she visited a hospital for headache. She also started complaining about difficulty in speaking and breathing after a while in the emergency room bed. She did not give any family history of atopy. However, she gave history of 2 episodes of similar swelling of the same side of the tongue within the last 9 months, both following intake of drugs for acid peptic disease (the drugs she couldn't recall). The symptoms in the past resolved after taking anti-histaminic that was recommended by a local hospital. She was also evaluated for joint pain one year back and had positive antinuclear antibody (ANA) and Double stranded DNA (DsDNA) and thus was diagnosed as systemic lupus erythematosus (SLE). She was not under any medications for this condition.

Her blood pressure in the ER was 110/80 mm of Hg, and pulse 120 bpm. She was afebrile and saturation was 92% in room air. Her systemic examination didn't yield any important information. She had her left half of tongue swollen and same sided swelling of floor of the mouth. The posterior pharynx was difficult to visualize. There was no wheeze in the chest and wheals on the skin. Her hemoglobin level was 11.8 gm% and her blood counts showed a total count of 8,100/mm<sup>3</sup> with 72% neutrophils, 25% lymphocytes and 3% eosinophils. The platelet count was 160,000/mm<sup>3</sup>. Peripheral blood smears showed normocytic normochromic RBCs with anisocytosis. Urine routine examination had 3+ albumin, and 15-20 pus cells. Her fasting blood glucose level was 137 mg/dl with normal renal function test and electrolytes.

She was managed in ER with injection Pheniramine 25 mg intravenous(iv), Inj. Hydrocortisone 100 mg iv stat and adrenaline nebulization and Inj. Adrenaline 0.05 mg IV. In ER itself she was able to talk, swallow and the difficulty in breathing was also relieved. The following day in the wards, the swelling had completely resolved.





(a) Before treatment (b) After treatment Figure 1. Unilateral swelling of tongue (a) before treatment and (b) after treatment

#### DISCUSSION

Patients with evidence of angioedema with risk of airway compromise, due to unknown etiology should be considered as histamine mediated edema and should receive epinephrine, steroids, antihistamines, and IV fluids. Epinephrine can be given via the intramuscular route or intravenous route. Intramuscular injection can be administered into the anterolateral middle third of the thigh, with initial dose 0.3–0.5 milligram (mg) of 1:1000 dilution (1 mg/mL), and if needed can be repeated every 5–20 minutes. IV epinephrine should be considered in patients requiring multiple doses of intramuscular epinephrine and should begin at doses of 1–4 micrograms (mcg) per minute.<sup>2</sup>

Antihistaminic and steroid can be used as adjuncts as they have slower onset of action. H1 antagonist like diphenhydramine can be used in doses of 25– 50 mg IV in combination with a second- or thirdgeneration antihistamine. Since 15% of cutaneous histaminic receptors are H2, the addition of an H2 antagonist is beneficial in decreasing urticaria. Steroids acts as anti-inflammatory mediators in histamine-mediated angioedema and anaphylaxis, typically taking 4-6 hours for its onset of action.<sup>2</sup>

In cases when there is suspect of non-histaminic medicated angioedema, mostly for hereditary angioedema (HAE) and ACE-inhibitors mediated angioedema, fresh frozen plasma (FFP), C1-INH concentrate, Ecallantide, a recombinant plasma inhibitor of kallikrein and Icatibant acetate, bradykinin B2 receptor antagonist are being used.<sup>2</sup> Ginseng is said to have anti-allergic effect but it is interesting to find that ginseng is associated with the unilateral angioedema in this patient. There is a case reported of 44-year male with rhinorrhea, respiratory difficulty with wheeze and abdominal pain 10 minutes after oral intake of fresh ginseng. The association of angioedema with ginseng intake was later confirmed by an oral challenge, a skin prick test (SPT), and a basophil activation test (BAT).6

There are conflicting research results for the incidence of allergic reactions in patients with SLE and without SLE ranging from 10% to 26% in patients with SLE compared to 4% to 20% in non

SLE patients.<sup>4</sup> Having SLE might have increased the risk of having recurrent allergic reaction to multiple drugs including Ginseng in this patient. However, we were not able to confirm the association with sensitization which is a drawback of this case study.

#### CONCLUSION

Allergic reactions present in different forms; hives, abdominal cramps, anaphylaxis and anaphylactic shock. It may also present as angioedema of the tongue. Unilateral swelling of tongue as presentation of angioedema is very unusual. Emergency physicians must be aware of this type of presentation of angioedema. Failure to recognize at earliest might result in failure to protect airways resulting in untimely death.

# Conflict of interest

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