Angioedema of the Left Maxillary Area

Joo-Wan Kang, Jong-Ho Lee, Jung-Heon Lee, Chi-Woong Song, Je Uk Park, Chang-Hyeon Kim

Department of Oral and Maxillofacial Surgery, Seoul St. Mary’s Hospital, Department of Oral and Maxillofacial Surgery, Yeouido St. Mary’s Hospital, The Catholic University of Korea School of Medicine

Abstract

Angioedema is defined as self-limited, localized swelling. The swelling is asymmetric, non-pitting, and non-tender. Common locations of swelling include periorbital area, lips, tongue, extremities, and bowel wall. A 54-year-old woman visited our hospital with the clinical complaint of left maxillary swelling. Swelling of the left maxillary area was diffuse and temperature of the involved area was normal. No infectious source was found on panoramic radiography and cone-beam computed tomography. After considering the patient’s medical history and assessing clinical examination, we suspected angioedema and administered corticosteroid only. In the oral and maxillofacial area, proper diagnosis and prompt treatment of angioedema is important because angioedema of the tongue or larynx may lead to airway obstruction or a life-threatening condition.

Key words: Angioedema, Corticosteroids

Introduction

Angioedema refers to abrupt and short-lived swelling of the skin, mucous membrane, or both[1]. It occurs in approximately 15% of the overall population and is more common in women than men[2]. Although the swelling is asymmetric, non-pitting, and non-tender, its effect can cause discomfort[3]. In addition, angioedema is characterized by recurrent attacks of local swelling in various parts of the body, particularly the face, oral cavity, pharynx, larynx, extremities, buttocks, and gastrointestinal tract[4]. Presentation of angioedema of the oral cavity varies, from minor edema to acute respiratory distress. The objective of this study was to report on a patient who had left maxillary swelling of an unknown origin and was treated with corticosteroid only.

Case Report

A 54-year-old woman (Fig. 1) with the clinical complaint of left maxillary swelling visited the Department of Oral and Maxillofacial Surgery, Seoul St. Mary’s Hospital, on June 1, 2011. The swelling had started spontaneously approximately two weeks ago and disappeared without treatment. However, the swelling showed recurrence one day before visiting our department. On clinical examination, there was a fixed induration around the anterior aspect of the left coronoid process, which measured approximately 1.5 cm. The patient had a history of surgery (canal up mastoidectomy, ossiculoplasty, and myringoplasty) for left chronic otitis media two months ago at the Department of Otorhinoparyngology of Seoul St. Mary’s Hospital. Another medical history was unremarkable and the patient
did not have any allergy to foods or drugs. On inquiry regarding past dental history, the patient had a severe swelling and inflammation reaction after extraction of the left maxillary first molar seven years ago. Panoramic radiography showed missing teeth of the left maxillary second premolar, first molar and both mandibular first and second molars (Fig. 2). However, there was no infectious source such as a periapical lesion or impacted teeth. For further evaluation, a cone-beam computed tomography (CT) scan was taken and only soft swelling of the left maxillary area was observed (Fig. 3). Clinically, we suspected an unspecified cellulitis and prescribed antibiotics (Moxicle tablet 375 mg, Amoxicillin and clavulanate; Daewoong, Seoul, Korea) for five days.

Two days later, the patient revisited Seoul St. Mary’s Hospital due to worsened facial swelling. The swelling
of the left maxillary area was diffuse, and no line of demarcation could be detected between normal and affected tissues. No pitting edema could be elicited on pressure. The temperature of the involved area was normal. The superior cervical lymph nodes were not palpable. Past medical history did not reveal any similar condition and there was no history of this condition among other family members. No evidence of infection was found anywhere on her body. As we suspected angioedema after considering all the factors, the patient was admitted to the

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**Fig. 4.** (A) Frontal photography five days after corticosteroid treatment. (B) Left facial photography five days after corticosteroid treatment. (C) Right facial photography five days after corticosteroid treatment.

**Fig. 5.** (A) Frontal photography 50 days after discharge. (B) Left facial photography 50 days after discharge. (C) Right facial photography 50 days after discharge.
Department of Oral and Maxillofacial Surgery for diagnosis and treatment. We started intravenous administration of corticosteroid (Dexamethasone disodium phosphate injection 5 mg; Huons, Seongnam, Korea) 10 mg per day for three days. To rule out infectious disease, complete blood cell count test and blood culture test were performed and the results of both tests were within normal limit. The patient’s vital signs were within normal limit and she did not experience difficulty with respiration during the hospitalization period. As the patient’s condition had improved and facial swelling had subsided, we reduced the corticosteroid dosage to 5 mg per day. After two days, the patient was discharged in better condition (Fig. 4).

After 50 days, on the return visit, the patient stated that she had not experienced facial swelling after discharge and the patient was in good health (Fig. 5).

This case report was approved by ethics committee of the medical school and informed consent was obtained from the patient.

Discussion

In 1876, Milton[5] clinically described the occurrence of a giant urticaria and named it angioedema. Angioedema is defined as self-limited, localized swelling[3]. Common locations of swelling include periorbital area, lips, tongue, extremities, and bowel wall[6], with rare involvement of the central nervous system[7]. Although angioedema is usually single, multiple lesions are occasionally observed[8,9]. Angioedema of the bowel wall may cause abdominal pain, nausea, and, rarely, bowel obstruction[10]. Angioedema of the tongue or larynx may cause airway obstruction[6] and often requires emergency treatment. The main cause of death from laryngeal edema is airway obstruction, with a mortality of 25% to 40%[11]. Therefore, determination of criteria that will enable identification of patients at risk of progressive airway compromise is important[12]. Episodes typically last between two and three days and may be isolated or recurrent[6].

The pathophysiology of angioedema consists of rapid onset of local increase in permeability of the submucosal or subcutaneous capillaries and postcapillary venules, causing local plasma extravasation and consequent ephemeral swelling[13]. There are a multitude of precipitating factors, temperature extremes, trauma, food sensitivity, and medications, such as penicillin and aspirin[14]. In recent years, the use of angiotensin-converting enzyme inhibitors (ACEI) has become the leading cause of angioedema. In some cases, psychological factors, such as emotional stress, appear to play an important role[7-9,15].

Although there are several different syndromes of angioedema, all have the characteristic swelling that reflects release of vasoactive mediators and transient increases in the permeability of postcapillary venules of subcutaneous and submucosal tissues[3]. The most frequent syndrome of angioedema is idiopathic recurrent angioedema, which occurs in 38% of patients[11]. In allergic angioedema, an identifiable allergen like foods or medications triggers a Type I hypersensitivity reaction in which activated mast cells release histamine and other vasoactive mediators[15]. Symptoms begin within minutes to 1 hour after exposure. Hereditary angioedema is an autosomal dominant disorder resulting from deficiency of C1 esterase inhibitor (C1 INH), which regulates the activity of the complement component C1 and has a regulatory role in the contact, fibrinolytic, and coagulation pathways[3]. Therefore, C1 INH deficiency results in unregulated activity of vasoactive mediators such as bradykinin, kallikrein, and plasmin. Acquired angioedema also results from C1 INH deficiency, which is thought to be autoimmune. Acquired angioedema usually presents after the fourth decade of life. Medication-induced angioedema occurs in 0.1% to 2.2% of patients taking angiotensin converting enzyme inhibitors and apparently occurs due to elevated levels of bradykinin[16]. Although the incidence is highest during the first month of taking the medication, the first event can occur after several years. Finally, cold, heat, vibration, trauma, emotional stress, and ultraviolet light can also induce angioedema, which is classified as physically induced angioedema.

For the diagnosis of angioedema, evaluation includes a detailed check of patient medical and familial history, physical examination, and laboratory investigation. Detailed patient history is required in order to identify potential triggers, such as allergens, medications, or trauma[16]. A physical examination is important in order to rule out other causes of edema, such as heart failure. In patients with a familial history of hereditary angioedema or with suspected C1 INH deficiency, serum markers of
complement activity should be assessed. As markers of mast cell degranulation are elevated during an acute episode of immunoglobulin E (IgE)-mediated angioedema, testing for allergen-specific IgE may be useful when an allergic trigger is suspected. In this case, after a thorough check of medical history and clinical examination, complete blood test and blood culture were performed and cone-beam CT was taken in order to assess the possibility of an infection. The absence of pain, heat, and redness differentiates this condition from an acute infective process. Therefore, after considering all of the factors, we suspected idiopathic angioedema.

The key to successful management of angioedema is detection and avoidance of triggers, early recognition of attacks, and aggressive airway management[16]. The first priority of acute management is airway protection. The absence of pain, heat, and redness of an infection. The absence of pain, heat, and redness differentiates this condition from an acute infective process. Although angioedema is common, it is not easy to differentiate from other infectious diseases. If angioedema is suspected, especially in the oral and maxillofacial area, proper diagnosis and prompt treatment is important in order to avoid development of life-threatening complications.

In idiopathic recurrent angioedema, glucocorticoid has been shown to be of some benefit in treatment; however, the risks of chronic therapy usually outweigh the benefits[3]. The patient should be reevaluated every three to four months in order to determine whether any new symptoms or identifiable triggers have developed in the interim. In this case, corticosteroid was administered as follows: 10 mg per day for three days, 5 mg per day for two days through the intravenous route. Five days after corticosteroid administration, the swelling had subsided and the patient had completely recovered.

Although angioedema is common, it is not easy to differentiate from other infectious diseases. If angioedema is suspected, especially in the oral and maxillofacial area, proper diagnosis and prompt treatment is important in order to avoid development of life-threatening complications.

References