

Angioneurotic edema: report of two cases

Chaitra .T.R.¹, Ravishankar .T.L.², Triveni Mohan Nalawade³

¹Senior Lecturer, Department of Pediatric and Preventive Dentistry, Kothiwal Dental College & Research centre, Kanth Road, Moradabad, India

²Reader, Department of Community Dentistry, Kothiwal Dental College & Research centre, Kanth Road, Moradabad, Uttar Pradesh, India

³Senior lecturer, Department of Pediatric Dentistry, Mannubhai Patel Dental College, Vadodara, India

Abstract

Pediatric angioedema exhibits a different cause and clinical manifestations than does adult angioedema. Unlike angioedema in adults, pediatric angioedema is caused mostly due to food, followed by insect bites, infection and antibiotics. Reactions to insect stings, both allergic and toxic, are commonly seen in medical pediatric practice but uncommonly encountered by pediatric dentists. Here we present two cases of angioedema involving the face mainly in children who presented insect bite in the affected region. Treatment and case resolution are described.

Keywords: giant urticaria, angioneurotic edema, allergy, pruritis.

Introduction

Angioedema is a rather common form of edema occurring in both hereditary and non-hereditary form. It appears to be closely related to general urticaria¹. Angioedema denotes similar but larger swellings of the deep dermal, subcutaneous and submucosal tissues². It is also referred to as “giant urticaria”, “Quincke’s edema” and also “angioneurotic edema”¹. Urticaria and angioedema are important components of systemic anaphylaxis which is an acute life threatening condition².

Reactions to insect stings, both allergic and toxic, are seen commonly in medical practice³, but uncommon in dental practice. Large local reactors are more frequent but rarely dangerous. The chance of a systemic reaction to an insect sting is low (5% to 10%) in large local reactors and in children with mild (cutaneous) systemic reactions.⁴ Herein, we present a 7-year-old boy and a 6-year-old boy with giant urticaria in whom the disease appeared due to insect bite on their forehead followed by swelling of face.

Case Report

Case 1

A 7-year-old boy reported to the Department of Pediatric and Preventive Dentistry, with diffuse facial swelling involving forehead and both the eyes (Figure 1). His history of present illness revealed, a millipede bite on the forehead one day before was the cause of initial swelling that involved the eyelid and gradually increased over 2 h to involve the entire face. The swelling remained throughout the day with no diurnal variations and no history of fever. General examination revealed normal vital signs and the boy weighed about 18 kg. On clinical examination, a diffuse swelling involving the whole of the forehead, upper and lower eyelid with bridge of the nose, right and left cheek region, was seen. The edema was non-pitting in nature with no local rise of temperature. Swelling was

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Correspondence to:
Chaitra T.R.

Department of Pedodontics and
Preventive Dentistry
Kothiwal Dental College & Research Centre
Kanth Road, Moradabad-244001
Uttar Pradesh, India
E-mail: chaitu4363@yahoo.co.in



Fig. 1. A 7-year-old boy exhibiting diffuse facial swelling involving forehead and both the eyes.

tender on palpation and there was no pus or serous discharge. Bite mark of the millipede was seen on the right forehead region (Figure 2). Intraoral examination revealed generalized dental fluorosis and initial caries lesions in teeth 16, 26, 36, and 46. Based on history and clinical features a provisional diagnosis of angioedema was made.



Fig. 2. Site of millipede bite on the forehead with diffuse swelling involving the entire forehead, upper and lower eyelid with bridge of the nose.

The patient was treated with combination of drugs. Injection of Avil (pheniramine maleate; 1cc IV) and hydrocortisone (100 mg IV) were given immediately (stat), which are believed to ameliorate delayed effects of anaphylactic reactions and may limit biphasic anaphylaxis. The combination of antihistamines and corticosteroids are routinely used in emergency treatment of anaphylaxis. Injection of Lasix (Furosemide; 20 mg IV stat) was given to

reduce the edema by reducing fluid retention and also attenuate the symptoms of troubled breathing. Tetanus toxoid injection was given to maintain active immunity as tetanus spores might have entered the body through insect bite. Injection of benzyl penicillin 7.5 lakh given three times daily (tid) empirically to prevent the bite area from secondary infection and also condition bear a resemblance to cellulites and Lyser D (combination of serratiopeptidase and diclofenac sodium) (tid) was given to reduce inflammation and alleviate the symptoms of pain. Tab Rantac (Ranitidine) 75 mg was given twice daily (bid). H_2 blocking drugs were used concurrently with H_1 antihistamine to reduce gastric secretion, and injection of Dexona (Dexamethasone; 1.5 cc tid) was also given. IV fluid DNS (dextrose normal saline) 10 drops/min was given. IV fluids were continued for 2 days. After 3 days, the edema reduced considerably with slight persisting in the lower eyelids and finally after a period of 8 days of intensive treatment, the swelling subsided completely (Figure 3).



Fig. 3. Recovery of the 7-year-old boy after 8 days.

Case 2

A 6-year-old boy reported to the Department of Pediatric and Preventive Dentistry with diffuse facial swelling mainly involving the philtrum area and the left lower eyelid region (Figure 4). The patient's parents suspected that the reason for the swelling was tooth decay and hence reported to our institution for check up.

History of the disease revealed that the swelling was sudden in onset and the parents reported immediately the next day morning. No episode attributable to food or drug allergy was present in history. Swelling had been diffuse,



Fig. 4. Six-year-old boy showing diffuse facial swelling mainly involving the philtrum area and the left lower eyelid region.

tender on palpation and non-pitting in nature with a small injury towards the left ala of nose, which was suspected to be an insect bite mark, but the patient's father did not reveal any history of insect bite or fever.

General examination revealed normal vital signs and the boy weighed about 15 kg. Intraoral examination showed no caries involvement of any tooth. Based on history and clinical examination, a provisional diagnosis of angioedema was made. It later responded to antihistaminic therapy.

Patient was treated with antihistaminic Allercet (Cetirizine; 5 mg once daily) and was asked to report in case of any aggravation of symptoms. Unfortunately the patient could not come for recall visit as he was from a distant place, but gratefully he called us up to let us know of his complete recovery.

Discussion

Clinics in previous decades predominantly consisted of children with allergic rhinitis, asthma and eczema, the proportion of children presenting with acute severe allergic reactions has progressively increased. Clinics are now dominated by acute food, insect venom and drug allergy, and there is evidence that these problems are increasing⁵. Angioedema is a variant of urticaria where there is involvement of the subcutaneous tissue⁶. Skin lesions may appear on the eyelids, lips, genitalia, tongue and pharynx. Sudden onset appears to be characteristic. Skin lesions last for few hours to few days⁷.

Angioedema affects males and females equally, usually during the 3rd and 4th decade of life. Pediatric angioedema exhibits a different cause, severity and clinical manifestations

than does adult angioedema⁸. Probable causes of angioedema are food (40%), insect bites (30%), infection (20%) and antibiotics (10%)⁵.

Reactions to insect stings, both allergic and toxic are seen commonly in pediatric practice³. Papular urticaria occurs through the bite of insects most commonly 'mosquitos' and bed bugs. On the contrary bee or wasps stings may produce severe acute urticaria or anaphylaxis, which may be life-threatening conditions. Hereditary angioedema usually manifests during childhood⁵ but it is rare and is transmitted as an autosomal dominant trait. Another common form occurring in young children is allergy to food including multiple food allergies, allergy to nuts and cow's milk^{2,5,8}.

In case 1, angioedema affected a 7-year-old boy with major manifestations on the face involving forehead and eyelids due to a millipede bite, which, to the best of our knowledge, is the first of its kind to be published. In case 2, angioedema affected a 6-year-old boy with mild manifestations on left side of the face, which responded to antihistamine therapy. No history of insect bite could be elicited; which is common⁹. The main difficulties arise around a lack of agreed definition for what constitutes an anaphylactic reaction. However, this then excludes patients who have developed angioedema and urticaria, which is appropriately treated prior to its evolution into a more severe and potentially life-threatening reaction. Conversely, if patients presenting just with angioedema and urticaria are incorporated into the definition, then this will include many individuals who will not have any progression of symptoms⁵.

Typical reactions following an insect sting include erythema, itching, pain, swelling and indurations localized to the area of the sting. These local reactions usually last only several hours and may respond to application of cool compresses. Large local reactions also occur frequently involving more extensive areas of the skin typically with swelling 5-10 cm in diameter (sometimes more) that is contiguous to the site of the sting. The swelling generally reach its peak in 24-48 h, but reactions can last up to 7-10 days.

Children exhibited predominantly facial (80 %) and lip (40%) edema. Although systemic reactions to insect stings are exception (less than 1% occurrences in children), they can be life-threatening conditions³.

Angioedema may present as an emergency and needs to be treated aggressively. Understanding the various possible causes is the first step in assessing angioedema. Allergic and drug-induced angioedema responds to removal of cause.² a stinger that remains in the skin after an insect sting should be removed. The area should be cleansed with soap and water. The immediate management of systemic reactions focuses on the treatment of anaphylaxis, for which administration of subcutaneous epinephrine (0.3 mL of a 1:1,000 dilution) is the treatment of choice^{3,8}. It is primarily used in the emergency treatment of non-hereditary angioedema involving larynx, it can be injected intramuscularly, subcutaneously or inhaled depending upon the severity of the reaction².

An antihistamine, such as diphenhydramine (Benadryl), or hydroxyzine (Atarax, Vistaril) may be given after

epinephrine has been administered to reduce pruritus and inflammation^{3,8}. When the conventional H1 and H2 antihistamine failed, other drugs like nifedipine is used as an adjunct to antihistamines. In resistant cases a brief course of systemic corticosteroids should be avoided because of significant adverse effects. Topical steroids also have been found to be effective². Some authors suggest the use of corticosteroids depending on clinical presentation³ while others mention corticosteroid therapy using intravenous dexamethasone sodium phosphate or hydrocortisone remains the main treatment for angioedema. Intravenous fluids for intravascular volume repletion and diuresis or ventilatory support for treatment of pulmonary edema have also been used⁸. Beta-agonists, oxygen histamine₂ blockers and vasopressors may also be useful depending on the clinical presentation³.

After 8 days of intensive drug therapy, the first patient described in this paper showed complete reduction in facial swelling and pain and was discharged.

Reactions to insect stings are seen commonly in pediatric practice, but seldom in pediatric dentistry practice. However, in some cases, history of insect bite cannot be elicited, as observed in our young patients. In addition, facial swellings similar to that presented in these cases also occur due to space infections following tooth decay. In such diagnostic dilemmas, it is our responsibility as health professionals to rule out dental causes and give appropriate treatment or refer the patients to pediatricians. As angioedema can also manifest fatally, dentists need to know, diagnose and treat such rare conditions promptly as "The eyes do not see what the mind does not know!!"

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