

Case Report

ANGIONEUROTIC EDEMA TRIGGERED BY PERICORONITIS: REPORT OF A CASE

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يعرض هذا التقرير حالة غير مألوفة لحدوث وذمة دموية ناكسة، ونعتقد أنها مسببة عن التهاب حاد للأنسجة المحيطة بتاج الرحى الثالثة السفلية (خرس العقل السفلي) البازغة جزئياً. وأثناء الهجوم الالتهابية الحادة بالإضافة إلى حدوث التهاب خلوي وجهي ثنائي الجانب، نرى حدوث وذمة في اليدين والأقدام. بينما في الهجوم الالتهابية الثانية الحادثة بعد ستة أشهر تقريبا نرى فقط التهاب خلوي وجهي ثنائي الجانب.

An unusual case of recurrent angioneurotic edema triggered, we believe on two occasions, by an acute pericoronitis related to partially erupted mandibular wisdom teeth is presented. During the first attack, in addition to massive bilateral facial cellulitis, edema of both hands and feet was seen. Whereas, in the second attack, about six months later, only bilateral facial cellulitis was the presenting feature.

The report suggests the possibility of pericoronitis triggering an attack of angioneurotic edema.

Introduction

Angioneurotic edema presents itself in two different forms, hereditary and non- hereditary. The neurotic implication has persisted since 1884 when the condition was first described as a condition which relates to psychological problems.¹ Later different etiological factors were implicated such as food or drug allergy, an endocrine disturbance or a focal infection.²⁻⁸ However, to date the etiology in most instances is unknown.

Angioneurotic edema is one of the immunologically mediated diseases manifesting itself as a clinical type of atopic allergy. It is characterized by episodic rapid swelling of respiratory tract mucosa, abdominal viscera, face, and extremities. The swelling usually lasts for 12-72 hours. The absence of heat and erythema differentiates the condition from infectious and inflammatory processes. The swellings are poorly defined and the edema is usually non-pitting, non-pruritic, painless, and involves the dermis and layers associated with it.¹⁻²

Angioedema can be classified into two clinical entities. First, the acute non- hereditary

angioedema characterized by a rapidly developing edematous swelling, mainly in the head and neck region, without any hematologic or immunologic pathological findings.^{3,9} Second is the hereditary angioedema, which can be defined genetically and biochemically as an autosomal dominant genetic disorder. In this case, the function of the serum protein C1 inhibitor is markedly reduced.^{3,9} Two forms of this type of edema have been described, the first being the common type (85%) where the concentration of C1 esterase inhibitor ranges from 0-50% Of the normal concentration with a mean level of 15%. The second variant occurs in 15% of the cases, and there is normal or elevated level of C1 esterase inhibitor. This is said to be an indication of functional deficiency.^{3,9}

Case Report

On 24 March 1987, a 27-year-old Sudanese lady presented at the Emergency Clinic of King Saud University College of Dentistry with a puffy face, both eyes closed, upper lip almost five times larger than the normal size while the lower lip was two times its normal size and both hands and feet were also swollen. The patient had a history of severe dental pain related to the lower left third

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molar the previous night followed by swelling within two hours. It started on the left side of the face, then increased during the next nine hours.

The patient denied using any self-medication, special food, cosmetics or having been bitten by an insect the night before. There was no history of recent trauma or previous similar swelling. There was no significant findings in the past medical or family history. On extraoral examination, non-pitting massive facial edema was observed. The eyes were almost closed, lips swollen [Figs. 1a, 1b] and both hands and feet were also swollen.

Intraoral examination revealed an inflamed pericoronal operculum related to the partially erupted left mandibular third molar [Fig. 2]. Hematological studies were performed and revealed no abnormal findings. Radiographic examination revealed bilaterally partially erupting lower third molars. The differential diagnosis included facial cellulitis, acute pericoronal infection, allergic reaction, hereditary angioneurotic edema, and acute angioneurotic edema triggered by pericoronitis. The absence of findings such as hotness, erythema, trismus, regional lymphadenopathy involvement, history of new medication, and history of similar swelling in the family rule out all the former diagnoses and confirm the diagnosis of acute angioneurotic edema triggered by pericoronitis, mainly because of the rapid episodic attack associated with inflamed pericoronal tissues.

Treatment:

The case was diagnosed as an angioedema of the acute type which was triggered by the pericoronitis. The patient was placed on phenergan 25 mg every 6 hours for 5 days; ibuprofen 200 mg for 5 days and Vitamin C 100 mg daily.

The patient improved remarkably under the treatment, and within the first day, the swelling subsided by more than 50%. This continued to regress in the subsequent days, and by the third day the swelling had completely disappeared [Fig. 3],

On April 5, 1987 the mandibular left third molar was extracted using 2.2 ml of 2% xylocaine with adrenalin 1:80,000 concentration. Follow-up for three months showed no recurrence of the symptoms. The patient was advised to have the right mandibular third molar tooth extracted but she refused.

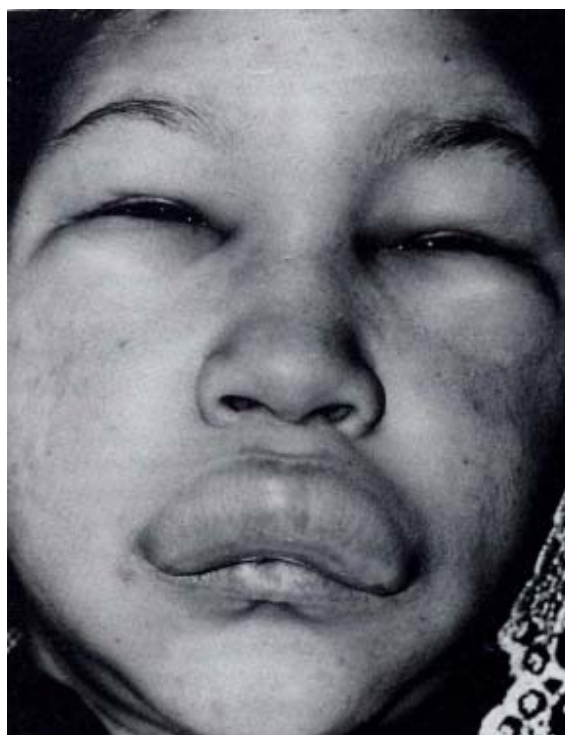


Figure 1a. Frontal view of the patient on the day of presentation (first attack). Note massive swelling of upper lip and face extending to lower eyelids.



Figure 1 b. Profile view of the patient on the day of presentation (first attack).



Figure 2. Intra-oral view (first attack) showing inflamed operculum related to the lower left third molar tooth.



Figure 3. Frontal view of the patient on the third day after treatment (first attack).

Second Attack

On November 7, 1987 the patient presented again with swelling, this time, on the right side of the face giving a similar history as on the previous occasion. Pain was experienced on the lower right third molar which started the night before and swelling followed after 3 hours. It only involved the right side of the face with the upper lip about 3-4 times the normal size. Hands and feet were not involved. Intraoral examination revealed an inflamed operculum related to the lower right

third molar. Again, there was no history of using unfamiliar medication, food, or cosmetics.

The case was then referred to an immunologist and dermatologist for consultation. Serological tests were performed and immunologic assay revealed normal levels of C2 (normal 0.03 g/l), C4 (normal 0.2 - 0.4 g/l) which further supports the diagnosis of acute angioedema. C2 and C4 levels were tested to detect any alteration and lack of the regulatory function of C1 inhibitor, which will consequently cause excessive cleavage of C2 and C4 and a detectable low level of C2, C4.^{3,4,9} Low levels of C2, C4 are therefore diagnostic for hereditary angioedema.¹¹ Similarly, normal levels of C2, C4 can confirm the diagnosis of acute angioedema in a similar way as there is a normal level of C1 inhibitor. Both serology and immunology consultants recommended keeping the patient under the same drug regime as in the previous treatment, together with corticosteroids.

Treatment:

The patient was treated similarly as on the previous occasion with the addition of steroids (Dexamethasone 8 mg I.V. Stat.). The swelling subsided completely within two days. One week later, the mandibular right third molar tooth was extracted using 2% Xylocaine, with epinephrine 1:80,000 concentration.

Prognosis:

Since November 1987, and until the present, the patient has been kept under observation and no further attack has been encountered.

Discussion

Although angioedema is a rare disorder, it can still be of great clinical significance to the dental practitioner. It carries a high morbidity and mortality rate and must be appropriately treated. Various drugs and therapeutic protocols have been used therapeutically and prophylactically for this condition.⁴⁻¹⁶

The prophylactic therapy of angioedema patients, prior to dental or oral surgery, includes administration of fibrinolytic agent, hormones and/ or fresh plasma 2-3 days before surgery,⁴⁻¹⁶ then two units of fresh frozen plasma (2 hours) before the surgery.¹³

Management of angioneurotic edema cases needs special care to manipulate the progressive airway obstruction which might only be achieved through hospitalization,⁴ nasotracheal or endotracheal intubation⁷ or tracheostomy.⁸

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