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Case Report

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Omalizumab in recurring larynx angioedema: a case report

Ayse Bilge Ozturk[1,*] and Emek Kocaturk[2]

[1]Adult Allergy Unit, Göztepe Tainning and Research Hospital, Istanbul Medeniyet University, Istanbul 34469, Turkey
[2]Department of Dermatology, Okmeydanı Trainning and Research Hospital, Istanbul 34126, Turkey

Angioedema with swelling of larynx is a serious allergic reaction and can be life-threatening. It can occur after exposure to various triggers and usually it is very difficult for the patient and the doctor to find the trigger and maintain complete remission. In idiopathic recurring angioedema presenting with frequent attacks, prophylaxis with H_1 antihistamines recommended. However, not all patients respond to antihistamines. Omalizumab is an anti-immunoglobulin (Ig)-E-Ig-G antibody approved for the treatment of asthma and also effective treatment in chronic spontaneous urticaria. We report a 47-year-old male patient with severe idiopathic angioedema controlled by corticosteroid and proggressed after discontining of corticosteroid because of its side effects. Omalizumab at a dose of 300 mg every 4 weeks was administrated and omalizumab provided a rapid clinical response after first injection. During the 4 months of omalizumab therapy, he had no further attacks and any other treatment needs. After 3 months of stopping omalizumab therapy, during the 4-week period he had two mild lip swelling in his lips that resolved with antihistamines.

Key words: Omalizumab, Angioedema, Larynx

INTRODUCTION

Recurring angioedema with the swelling of larynx can be life-threatening. It can occur after exposure to various triggers and usually it is very difficult to find the trigger and maintain complete remission. The antihistamines are the first line recommended therapy [1]. However, not all patients respond to antihistamines. Corticosteroids, epinephrine and immunosuppressive therapies are the other treatment options [1]. However corticosteroids and immunosuppressive therapies cannot be used for very longer due to their side effects. Here, we report a case of severe angioedema that responded to corticosteroid, initiated omalizumab due to corticosteroids' side effects and showed complete remission with omalizumab.

Correspondence: Ayse Bilge Ozturk
Adult Allergy Unit, Göztepe Tainning and Research Hospital, Istanbul Medeniyet University, Istanbul 34469, Turkey
Tel: +90 5434336679
Fax: +90 2165664065
E-mail: aysebilgeozturk@yahoo.com

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CASE REPORT

A 47-year-old man had symptoms of weekly recurrent angioedema attacks in lips, tongue, larynx, and scrotum for seven years and experienced swelling of larynx twice weekly in the last 3 months. He had been receiving antihistamines and the symptoms were relapsing during the antihistamine treatment. His signs and symptoms did not improve during administration of different nonsedating H_1-antihistamines or increasing of the standard dosage up to fourfold of the daily. Emergency treatment with prednisolone and short acting antihistamines led him to a remission in emergency department visits. Urticaria was absent. Physical examination was unremarkable. He had no other disease and did not receiving drugs such as angiotensin-converting-enzyme inhibitors. Any of the relatives had no angioedema. Laboratory findings included normal values for complete blood count with differential, comprehensive metabolic panel including antinuclear antibody, thyroid stimulating hormone level, C4 level, C1 inhibitor function and level and total immunoglobulin (Ig) E level of 183 kU/L. Also C4 levels were all in normal range during the angioedema attacks. The patient did not attribute his angioedema to certain foods. But the stress is a trigger for him. Fluoksetine and 0.5 mg/kg prednisolon therapy was initiated and clemastine was recommended for the emergency new attacks during corticosteroid therapy. Soon after corticosteroid therapy patient's angioedema regressed. However he felt more depressed after the therapy and the treatment started to be tapered for this side effect. After reducing the corticosteroid dose angioedema attacks again progressed. Because of the patient's angioedema, 300 mg of omalizumab every 4 weeks administered subcutaneously after obtaining written informed consent. His last episode of angioedema was seen 3 days before initiation of therapy. He has tolerated tapering and cessation of his oral steroids and he has remained free of angioedema during 4 months of omalizumab therapy. After 3 months of stopping omalizumab therapy, during the 4-week period he had two mild lip swelling in his lips that resolved with antihistamines.

DISCUSSION

Omalizumab is an anti-immunoglobulin (Ig)-E-Ig-G antibody approved for the treatment of asthma and the studies have showed that omalizumab is also effective for antihistamine-resistant chronic urticaria [2, 3]. There are case reports in limited number reporting antihistamine refractory idiopathic angioedema that resolved on treatment with omalizumab [4, 5]. First, Sands et al. [4], reported 3 angioedema cases. In this report two of the patients had asthma and omalizumab was also used for their asthma. The drug dose was adjusted according to disease severity and IgE level and symptoms were improved after the therapy. Later, von Websky et al. [5], presented a 68-year-old man who had weekly recurrent severe angioedema attacks and had failed to respond to antihistamines. A 300-mg omalizumab was administred every four week and after the first injection, the patient still had mild angioedema. Similarly, we used the same dose and our clinical response was more rapid. Hypothetically, omalizumab could be also effective in idiopathic angioedema with the same mechanism in antihistamine resistant chronic urticaria.

In conculusion, omalizumab provides a rapid clinical response with less side effects in severe idiopathic angioedema without urticaria.

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