



RESEARCH ARTICLE

SEVERE AND RECALCITRANT CHRONIC URTICARIA RESOLVED AFTER FILLER REMOVAL.

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Abstract

Chronic idiopathic urticaria is a common skin disease without a clear etiology in the vast majority of cases. The main therapeutic options are directed towards histamine receptors. When antihistamines failed, other therapeutic options may be added such as systemic steroids, mast cell stabilizer and more recently Omalizumab. Herein we report a rare case of severe and recalcitrant urticaria that was resistant to many treatments modalities including the above-mentioned drugs. Extensive patient history led to two possible triggers, which are lip fillers and tooth filling. Removal of both fillings cured her disease completely. Dermatologists should be aware of any new procedures and materials administered to patients prior to development of urticaria.

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Introduction:-

Urticaria for more than six weeks is considered chronic and in majority of cases more (80%) no exogenous causes can be determined. The term chronic idiopathic urticaria (CIU) [1,2,3] is commonly used. Antihistamines, systemic corticosteroids, mast cell stabilizers and biological therapy (omalizumab) are being used. Dermatologists often exposed to unresponsive cases to several treatment modalities. In this paper we are reporting a patient with severe and recalcitrant chronic urticaria for more than 7 months that is only completely resolved after removal of lip filler and tooth filling.

Case Report:-

46 years old female, medically free, presented to our clinic with history of severe urticaria for 7 months. The rash started to appear in the face with mild facial angioedema that progressed as generalized urticaria all over the body. The patient started by herself on Cetirizine 10 mg for two weeks, without benefit. Later on she was prescribed Hydroxyzine 25 mg and loratidine 10 mg. Minimal response was noted and after two weeks she was started on Prednisolone 40 mg in addition to the antihistamines. Up to 50% of the rash has improved over the body but angioedema over the face continued to appear. Montelukast 10 mg and doubling the dosage antihistamines brought only little benefit. Omalizumab injection was initiated every week at 300 mg that brought good benefit along with corticosteroid. Corticosteroid was tapered down till 10 mg but patient stopped it completely because of weight gain. Urticaria flared up very significantly with Angioedema of the face (while she still on Omalizumab). Prednisolone 40 mg was taken by the patient irregularly for social occasions to suppress the hives. At this stage, she sought medical advice with other dermatologist. Detailed history revealed that patient has both tooth filling and lip filler (JUVEDERM VOLIFT, Allergan, Inc. Irvine, California, USA) have been administered for her 2 and 1 months

prior to urticaria onset respectively. Patient was advised to remove lip filler first and in one month the tooth filling. Hyaluronidase was injected twice (three days apart) to dissolve the lip filler. However, within few days patient chose to remove the tooth filling by her dentist against our advice and not to wait for a month. In less than one week she noticed significant decrease in number of hives that completely disappeared in a month. She is off all treatments without any recurrence for more than six months.

Discussion: -

None of the theories of pathogenesis of chronic urticaria (CU) has been fully established⁴. The best-developed hypotheses include the autoimmune theory, theories involving histamine-releasing factors, and the cellular defects theories⁵. IgE-mediated, cell-mediated and complement-mediated were established as main pathogenesis¹. Dermatologists commonly failed to find out exogenous causes only in 20 to 30% they could be determined. In the literatures, unusual triggers had been reported such as hidden infection including *Helicobacter pylori*⁶, Hepatitis A⁷ and Hepatitis C⁸, eye drops and tooth filling have been reported.^{9,10} Local skin reaction to Hyaluronic acid at site of injection is well known with all types of fillers that can be confusing as angioedema. In two reports, angioedema affecting lips post fillers has been described, however in the first case¹¹ it seems to be an overt reaction to local filler rather than a classic immune-mediated angioedema. It occurs within minutes at the site of injection only and disappeared in few days. In the second one¹² Herpes Simplex Virus has been associated with the swelling. In both papers filler hasn't been dissolved and no recurrence has been reported. We expected in those patients that urticaria should continue to flare as long as fillers still in the body.

In contrast to our patient, generalized urticaria has happened within a three to four weeks of injection and only cleared after dissolving lip filler and removal of tooth filling. Another case of urticarial vasculitis (approved by skin biopsy) has been reported¹³. Three weeks after filler and resolved within six weeks later. However in this case filler hasn't been dissolved with hyaluronidase, which might doubt for the possible relationship to cause urticarial vasculitis. Another report of angioedema that related to hyaluronidase rather than lip filler¹⁴. The possibility that the angioedema in this patient was related to the dissolved filler rather than only hyaluronidase cannot be ruled out. The antigen exposure to the immune system following degradation by hyaluronidase is a possible pathogenesis.

In our patient, both lip filler and tooth filling were administered 1 and 2 months respectively prior to her symptoms. No clue to us which of them is the true culprit. Although we advised her to remove them one month apart, she decided removing them simultaneously, which creates uncertainty, which is the main culprit. Complete remission of urticaria within few weeks of removal of both fillings creates strong evidence for the cause-relationship. Nevertheless, the message from this patient is the importance of detailed and extensive history in finding the possible causes and relieving the frustration and safe huge cost.

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