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Case Report

Facial angioedema in young elderly: Rare complication of rare drug

Saniya Khan^{1,*}, Sunil Kumar¹, Dhruv Talwar¹, Akhilesh Annadata¹, Ayush Dubey¹

¹Dept. of Medicine, Jawaharlal Nehru Medical College, Datta Meghe Institue of Medical Sciences and Research (Deemed to be University), Sawnagi Meghe, Wardha, Maharastra, India



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ABSTRACT

Angioedema is an intense often disfiguring but temporary swelling of a localized area which usually involves superficially lying structures like skin, mucosa along with subcutaneous tissues. Areas which are often affected by angioedema have a wide range starting from the face, lips, tongue, pharynx, the supraglottic area and rarely, the subglottic area. It might also effect the gastrointestinal mucous membranes, genitalia, hands and feet. There maybe variety of causes for development of angioedema including various drug intake, insect bites or stings and certain types of food products particularly nuts, milk and eggs. Levamisole is known to have anthelminthic and immunomodulatory properties and is a common contaminant in cocaine due to its mood stimulatory action. Here we report a case of a 60-year-old female who presented with angioedema following ingestion of a drug known as levamisole for its anthelminthic activity.

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1. Introduction

Angioedema is a abrupt onset swelling having short duration involving the skin as well as the mucous membranes. Although a wide range of parts of body may be involved but the swelling most commonly arises around the eyes and lips. Angioedema can vary from a benign disease to a life threatening swelling involving the respiratory tract and intestines. Acute pain in abdomen or recurrent chronic pain of the abdomen can be rare presenting symtoms. This pain in the abdominal is often described by the patient to be of cramping or colicky type. Levamisole is a synthetic imidazothiazole derived drug which has antihelmenthic properties along with immunomodulatory functions. ¹

In the past levamisole was used to treat inflammatory conditions such as rheumatoid arthritis, crohn's disease and nephrotic syndrome. Common side effects include neutropenia, agranulocytosis as well as vasculitis. Rarely more severe side effects ranging from leukoencephalopathy,

E-mail address: Saniya.24@hotmail.com (S. Khan).

arthritis, type 1 and 3 hypersensitivity reactions, ulcers of mouth and skin to various gastrointestinal symptoms might be seen.³

In the current scenario, exposure to levamisole occurs most often along with cocaine which is contaminated with levamisole as levamisole also as mood stimulatory effects. Most common presentation following consumption of cocaine contaminated with levamisole is seen as a tender, purpuric rash in retiform distribution with erythematous borders and necrotic centers. Lesions have predominance towards the lower limb. We report a rare case of levamisole induced angioedema in a 60-year-old female who had ingested levamisole in view of its anthelminthic activity.

2. Case Report

A 60-year old female patient presented with complaints of swelling of face including the eyes and lips and rashes over bilateral upper limbs and back since 2 days. (Figure 1). There was no history of difficulty in breathing, itching, headache, fever, cold, cough, palpitations, chest pain.

^{*} Corresponding author.

No h/o oral or nasal regurgitation

There was no any sensory involvement. Patient had a history of intake of Tablet levamisole for 2 days prior to her onset of symptoms which she was taking as a anti helminthic agent



Fig. 1: Showing swelling of face

No history of fever, jaundice, joint pains or rashes. On general examination patient had pulse of 98/min and blood pressure was 100/80mmhg. Patient was adequately built. Patient had facialpuffiness with swollen eyes and lips. Red rashes were found on the dorsal aspect of the forearm and back which were not associated with itching.

Laboratory investigations revealed following values: Haemoglobin of 7.5 gm%, MCV of 70 and white blood cell count of 14000 per cumm with absolute eosinophil count of 800 cells/microlitre. Serum Albumin 3.9 gm per dL, Total serum protein 7.1 gm per dL, Aspartate transferase and Alanine transferase were 27 and 21 IU/L respectively, urea 25 mg per dL, creatinine 0.6 mg per dL, serum Na⁺ and K⁺ were 140 and 3.8 meq per L respectively.

Patient was immediately admitted in ICU and started on injectable steroids along with anti histaminics. Tablet levamisole was discontinued. During the course of hospital stay patient improved clinically and her swelling reduced significantly. She was discharged 5 days later and is doing well on follow up.

3. Discussion

Pathophysiology of Angiodema is due to a quick increase in the permeability of submucosal or subcutaneous capillaries and post capillary venules with extravasation of plasma locally. Most causes of angioedema are based on release of histamine or bradykinin however other substances that are vasoactive might be contributory.

Release of Histamine can occur due to antigen dependent cross linking with immunoglobulin E to the surface of mast cells or basophils forming a cascade of allergic reaction. However, Activation of these same cells can also occur due to auto immune pathology by formation of antibodies that are IgG antibody against IgE or IgG. Angiodema accompanied with urticaria is caused due to release of histamine. Angiodema is most commonly seen with urticaria and rarely without it.

Bradykinin is the key modulator of angioedema which is in association with angiotensin-convertase enzyme inhibitor (ACE Inhibitor) which inhibit the destruction of Bradykinin hence increasing its concentration in the body. Bradykinin is formed from plasma or tissue-bradykinin forming pathway.

Swelling that is associated with angioedema are often symmetrical facial or puffiness of hands or peripheral pitting edema or persistent facial swelling.

Levamisole, is a drug which is an anthelmintic and it's use is associated with neutropenia, agranulocytosis, vasculitis or vasculopathy associated with skin manifestations and can mimic certain autoimmune diseases. ⁵ Commonly seen a contaminent in cocaine levamisole has limited spectrum for use in today's time. ⁶ In our unique case we have reported a case of a middle aged female who had suspicion of helminthic infection for which she had taken levamisole and had presented with angioedema with swelling of face and around eyes which subsided later with steroids and anti-histaminic.

4. Conclusion

In this case report a drug induced angioedema lead to presentation with swelling over the face along with rashes and it responded well to steroids and anti histaminics. To conclude, this case highlights one of the rare drugs such as levamisole manifesting with an adverse reaction like angioedema which with prompt diagnosis and careful selection of management was manageable with Intravenous steroids.

5. Source of Funding

None

6. Conflict of Interest

None.

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Author biography

Saniya Khan, Post Graduate Resident

Sunil Kumar, Professor

Dhruv Talwar, Post Graduate Resident

Akhilesh Annadata, Post Graduate Resident

Ayush Dubey, Post Graduate Resident

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