Short Communication

Two varied presentations of cold urticaria

Sir, we report two cases of cold urticaria, who were confirmed on rechallenge.

Patient 1 A 20-year-old female presented with itching and wheals on the face and legs of 6 months duration. There was no history of development of similar lesions on other parts of the body. An extensive history taking revealed that patient developed lesions only while attending the air-conditioned classrooms or the centrally air-conditioned library. There was no history of atopy, angioedema, systemic symptoms or intake of any medicines. Neither wheals nor any clinical abnormality were detected on clinical examination. The ice cube test performed on the patient was positive (**Figure 1**).

Patient 2 A 14-year-old male came with complaints of wheals and itching only on face and upper extremities of 2 month duration. There was no history of development of similar lesions on other parts of the body. No history of aggravation of lesions on sun exposure was noted. A detailed history revealed the development of symptoms only while walking through the fields in the early hours of morning with the temperature ranging from 16 to 20°C. There was no development of wheals while walking through the fields in the noon or evenings. There was no history of atopy, angioedema, systemic symptoms or intake of any medicines. The lesions had occurred only after the onset of the cold seasons. On the basis of his history, a diagnosis of cold urticaria was suspected and an ice cube test was performed which was positive (**Figure 2**).

Figure 1 Positive ice cube test in patient one.

Figure 2 Positive ice cube test in patient two.

Discussion

Cold urticaria is a type of physical urticaria where exposure to cold leads to the formation of wheals. The incidence of cold urticaria has been reported to range from 9.5% to 20%.[1] Cold urticaria is broadly classified into primary (96%) and secondary (4%). Immediate cold contact urticaria is the commonest type of primary cold urticaria which is characterized by the appearance of wheals following minutes of exposure to cold.[2] The wheals last for about an hour or so. Cold winds and rain are stimuli for development of cold urticaria. Occasionally cold urticaria may be associated with systemic symptoms like flushing, palpitations etc.

The ice cube test or the cold contact test is the most common and standard test done for

evaluation of cold urticarial.[3] This is positive only in generalized and localized cold contact variants and it is negative in systemic cold urticaria and secondary cold urticarias.[1] In this test there is appearance of wheal on rewarming of the skin which has been exposed earlier to an ice cube for five minutes.[2] In order to detect the threshold cold temperature, various devices like Temp test® are present, but not easily available.[3]

The pathogenesis of primary cold urticaria is exactly not known but it has been proposed that there is production of IgE antibodies following an unknown antigenic stimulation. At low temperature, interaction between these antigens and IgE antibodies attached to mast cells occurs which leads to the activation of mast cells and subsequent release of mast cell mediators like histamine, prostaglandin D2, platelet activating factor, leukotrienes.[4,5]

Patients with cold urticaria present with wheals only in situations where there is exposure to cold. Our first patient presented with wheals only when sitting in air conditioned rooms whereas the second patient presented with wheals on walking in the fields only in the early hours of the morning. This stresses the most important fact that a proper history is the most important part in the management of urticaria, where avoidance of the trigger event can lead to subsidence of disease. Unless the cause is found the urticaria may become chronic leading to distress to the patient and the treating physician. We thus present 2 cases of cold urticaria from two different backgrounds- one from the urban setting and one from a rural area, where a proper history helped in diagnosis.

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Phobia from scarring: the invisible problem of cutaneous leishmaniasis in the endemic area

Sir, phobia (from the Greek: φόβος, Phobos, meaning "fear" or "morbid fear") is a type of anxiety disorder, usually defined as a persistent fear of an object or situation in which the sufferer commits to great lengths in avoiding despite the fear, typically disproportional to the actual danger posed, often being recognized as irrational.[1] A patient may have phobia from skin diseases or the treatment used for them. Phobia from systemic or topical steroids[2-4] is well known medical condition. Similarly skin diseases, like

hyperhidrosis, may generate social phobia.[5] In addition a dermatologist may encounter a patient who has a trypanophobia which is an extreme fear of medical procedures involving injections or hypodermic needles**.**[6]

In this brief letter, I want to share my observation of an 18 year-old-girl who had big ulcer in the nose, as an end result of her excessive fear from leishmaniasis scar.

The patient hailing from a village endemic for cutaneous leishmaniasis (CL), was thought to have an acne lesion in her nose. She was afraid of her face getting scarred as she was still not married. She was thinking that there was no effective medical treatment available for this disease and scarring was inevitable. She sought advice from a traditional therapist who did aggressive shaving of the lesion that ended by producing a large ulcer over the nose which eventually required skin grafting.

Leishmaniasis, as seen in Saudi Arabia, has been a major cause of scarring of the face which has given rise to a fear among the people. Indigenous practitioners by mismanagement add fuel to the fire by endorsing this misconception amongst the people. This case also highlights the importance of understanding the perceptions of the patients toward skin diseases, particularly in adolescents where cosmetic appearance is of paramount.

I believe that public health workers should reassure the community that effective treatment is available for CL. On the other hand patients' education, in the media, about diseases like CL, should be properly disseminated to avoid both fear and improper advice leading to complications as exemplified.

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Angiolymphoid hyperplasia with eosinophilia

Sir, a 19-year-old boy presented with multiple nodules over postauricular region on right side since last 3 years. Lesions were asymptomatic. On examination nodules were found to be of variable sizes and were skin coloured (**Figure1**). Nodules were non-tender. No such other lesions are present anywhere on body. On systemic examination other systems were found to be normal including the reticuloendothelial system.

Figure 1 Multiple skin-coloured nodules over post auricular area.

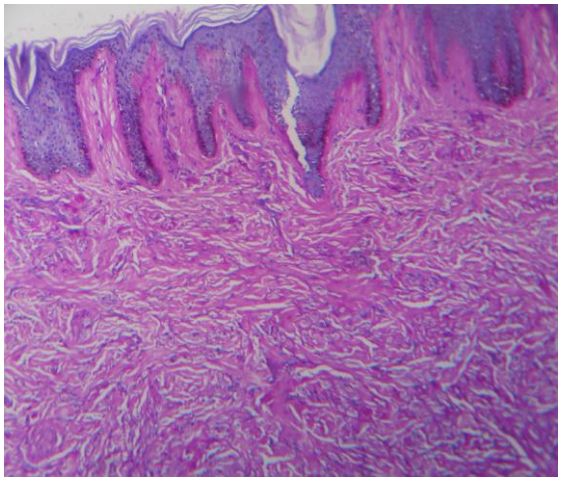

Figure 2 Plethora of blood vessels in dermis with plump endothelial cells in abundant collagenous stroma (H&Ex10).

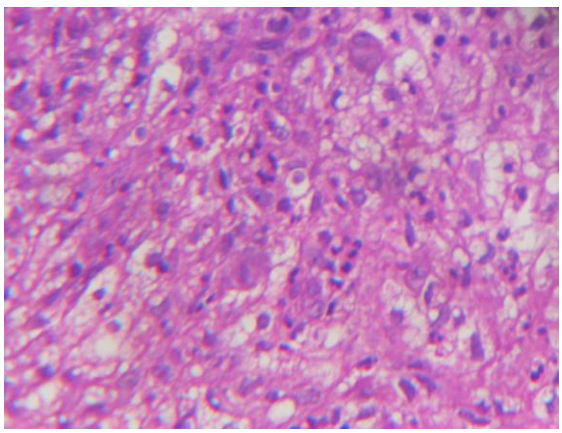

Figure 3 Portion of dermis showing endothelial cells with eosinophilic cytoplasm or vacuolated cytoplasm, some of which have invaded the lumen of the blood vessels (H&Ex100).

Complete blood count and ESR were normal and no eosinophilia was detected. Histopathological examination of skin biopsy specimen showed hyperkeratosis, acanthosis with plethora of blood vessels some in cord like distribution and others dilated with plump endothelial cells in abundant collagenous stroma of dermis (**Figure 2**). Dilated blood vessels were lined by plump (epithelioid) endothelial cells with eosinophilic cytoplasm or vacuolated cytoplasm, some of which had invaded the lumen of the blood vessels (**Figure3**). Some giant cells were also present.

Discussion

Angiolymphoid hyperplasia with eosinophilia, first described by Wells and Whimster in 1969, is a rare benign vascular tumor.[1] Both sexes are equally affected. Aetiology has not been identified and is thought to be a reactive process. Trauma, hormonal changes and infections (HTLV or HHV 8) have been suggested to play a role in the pathogenesis.[2] Patients usually present cluster of small translucent nodules on the head and neck. Other sites of the body like oral mucosa, extremities may be involved. Involvement of the deeper tissue, internal organs and bones can also occur. Contrary to as the name suggests, blood eosinophilia is not always present.[3]

Histopathological examination shows numerous proliferating blood vessels which are lined by endothelial cells. Lymphocytic and eosinophilic infiltrate may surround the vessels. Cytological atypia may rarely be seen.

Main differential diagnosis of angiolymphoid hyperplasia with eosinophilia is Kimura's disease. Kimura's disease is differentiated by occurrence in younger age group, presence of deeper-seated lesions, association of lymphadenopathy and absence of epithelioid cells in histopathlogy.[4] Exceptionally, both diseases may co-exist.[5]

Lesions usually regress spontaneously. Surgery and radiotherapy are also effective.

Treatment with Nd:YAG laser and imiquimod also shows promising result.[6]

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Angina bullosa hemorrhagica: an idiopathic intraoral blistering disorder

Sir, Badham coined the term 'angina bullosa hemorrhagica' to describe a condition of oral mucosa which is affected by blood filled painful blisters of abrupt onset. Though there are few reports mentioning several predisposing factors for this disease, but exact etiopathogenesis is still unclear. There is no specific investigation to reach a final diagnosis.

A 49-year-old woman, school teacher by profession, presented with gradually enlarging painful ulcer in the oral cavity for 5 days. (**Figure 1**) The ulcer was preceded by few hemorrhagic blisters, which developed after a prodrome of pain and burning sensation. The blisters developed on hard palate, and coalesced to form a large single bulla before rupturing. The bulla soon ruptured leaving erosion. She was having difficulty in chewing food, forcing her to survive on fluids only. She did not have similar blisters in other parts of the body. She had no history of similar illness in past or in her family members. There was no history of trauma prior to onset of lesions. She had no present or past history of any major bleeding episode. She did not mention any recent or long term medications for other illness or any kind of recent dental procedures.

Cutaneous examination revealed that she had dark reddish irregular linear erosion along the midline of the palate. Other mucocutaneous as well as systemic examination were non contributory. All laboratory parameters including coagulation profile were within normal limit. Clinical diagnosis of angina bullosa hemorrhagica was made. Patient was

Figure 1 An arciform ulcer on hard palate.

reassured and asked to come back after 7 days, prescribing only chlorhexidine mouth rinse and lesion healed within 7 days. As the lesion healed and did not recur, histopathological and immunological examinations were not done.

'Angina bullosa hemorrhagica', is an essentially benign condition affecting mucosa of oropharynx, characterized by sudden appearance of blood filled submucosal blisters of unknown aetiology. No blood dyscrasias, vesicobullous disorder or any systemic illness are associated. In 1933, it was first described as 'traumatic oral hemophlyctenosis (TOH)'. The term 'angina bullosa hemorrhagica (ABH)' was used by Badham for the same condition in 1967 and was later renamed as 'recurrent oral hemophlyctenosis (ROH)'.[1]

Discussion

ABH is a disease of middle-aged and elderly population, characterized by an acute onset of blood-filled vesicles or bullae, usually painful, commonly affecting mucosa at the junction of soft and hard palate. The buccal mucosa, lateral and ventral border of the tongue may be affected; however, masticatory mucosa is usually spared.[2] Initially there is painful, dark red, intact bulla which ruptures in few hours leaving a bleeding, painless ulcer with spontaneous healing within two weeks without any scar. Some patients may correlate these bleeding episodes with meals or a prodrome of burning sensation.[3] Though there are no definite etiological factors, but diabetes, inhaled long-term steroids, trauma, restorative dentistry, periodontal therapy, hereditary predisposition, found to have variable degree of association, without any hematological or immune-pathological disorders in background.[1,2,4-6]

In histopathology, there is subepithelial separation from lamina propria, mild inflammatory infiltrate and parakeratosis. Immunofluorescence study has failed to demonstrate any immunoglobulin or fibrin deposits along basement membrane zone.[4]

ABH has to be differentiated from blood dyscrasias (thrombocytopenia, von Willebrand's disease, other coagulopathies and leukemia), immunobullous disorders (bullous pemphigoid, linear IgA disease, cicatricial pemphigoid, and dermatitis herpetiformis), angiomatoid lesions in Osler-Rendu-Weber syndrome. ABH is a clinical diagnosis, but thorough history, clinical examination, complete hemogram, coagulation profile, histopathological examination and immunofluorescence study are necessary to exclude these conditions.

As it is a benign disorder, no treatment is required except for large pharyngeal bullae which may necessitate tracheostomy. Sometimes NSAIDs or antimicrobials as rinse and systemic therapy is given for symptomatic relief and to prevent secondary infections. Lastly, patient needs counselling for regular follow up to exclude those mimicking conditions properly.[7]

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