EDITORIAL
Management of autoimmune urticaria
Arun C. Inamadar, Aparna Palit ................................................................. 89

VIEWPOINT
Cosmetic dermatology versus cosmetology: A misnomer in need of urgent correction
Shyam B. Verma, Zoe D. Draelos ................................................................. 92

REVIEW ARTICLE
Psoriasiform dermatoses
Virendra N. Sehgal, Sunil Dogra, Govind Srivastava, Ashok K. Aggarwal .... 94

ORIGINAL ARTICLES
A study of allergen-specific IgE antibodies in Indian patients of atopic dermatitis
V. K. Somani .................................................................................................. 100

Chronic idiopathic urticaria: Comparison of clinical features
with positive autologous serum skin test
George Mamatha, C. Balachandran, Prabhu Smitha ..................................... 105

Autologous serum therapy in chronic urticaria: Old wine in a new bottle
A. K. Bajaj, Abir Saraswat, Amitabh Upadhyay, Rajetha Damisetty, Sandipan Dhar ................................................................. 109

Use of patch testing for identifying allergen causing chronic urticaria
Ashimav Deb Sharma .................................................................................. 114

Vitiligoid lichen sclerosus: A reappraisal
Venkat Ratnam Attili, Sasi Kiran Attili ......................................................... 118
BRIEF REPORTS

Activated charcoal and baking soda to reduce odor associated with extensive blistering disorders
Arun Chakrarthi, C. R. Srinivas, Anil C. Mathew .......................................................... 122

Nevus of Ota: A series of 15 cases
Shanmuga Sekar, Maria Kuruvila, Harsha S. Pai .......................................................... 125

Premature ovarian failure due to cyclophosphamide: A report of four cases in dermatology practice
Vikrant A. Saoji .................................................................................................................. 128

CASE REPORTS

Hand, foot and mouth disease in Nagpur
Vikrant A. Saoji .................................................................................................................. 133

Non-familial multiple keratoacanthomas in a 70 year-old long-term non-progressor HIV-seropositive man
Hemanta Kumar Kar, Sunil T. Sabhnani, R. K. Gautam, P. K. Sharma,
Kalpana Solanki, Meenakshi Bhardwaj ................................................................. 136

Late onset isotretinoin resistant acne conglobata in a patient with acromegaly
Kapil Jain, V. K. Jain, Kamal Aggarwal, Anu Bansal .................................................. 139

Familial dyskeratotic comedones
M. Sendhil Kumaran, Divya Appachu, Elizabeth Jayaseelan .................................. 142
Nasal NK/T cell lymphoma presenting as a lethal midline granuloma
Vandana Mehta, C. Balachandran, Sudha Bhat, V. Geetha, Donald Fernandes ................................. 145

Childhood sclerodermatomyositis with generalized morphea
Girishkumar R. Ambade, Rachita S. Dhurat, Nitin Lade, Hemangi R. Jerajani ................................. 148

Subcutaneous panniculitis-like T-cell cutaneous lymphoma
Avninder Singh, Joginder Kumar, Sujala Kapur, V. Ramesh .............................................................. 151

LETTERS TO EDITOR

Using a submersible pump to clean large areas of the body with antiseptics
C. R. Srinivas ................................................................................................................................................. 154

Peutz-Jeghers syndrome with prominent palmoplantar pigmentation

Stratum corneum findings as clues to histological diagnosis of pityriasis lichenoides chronica
Rajiv Joshi ...................................................................................................................................................... 156

Author’s reply
S. Pradeep Nair ............................................................................................................................................. 157

Omalizumab in severe chronic urticaria
K. V. Godse .................................................................................................................................................. 157

Hypothesis: The potential utility of topical eflornithine against cutaneous leishmaniasis
M. R. Namazi ................................................................................................................................................ 158

Nodular melanoma in a skin graft site scar
A. Gnaneshwar Rao, Kamal K. Jhamnani, Chandana Konda ................................................................. 159
Palatal involvement in lepromatous leprosy  
A. Gnaneshwar Rao, Chandana Konda, Kamal Jhamnani .............................. 161

Unilateral nevoid telangiectasia with no estrogen and progesterone receptors in a pediatric patient  
F. Sule Afsar, Ragip Ortac, Gulden Diniz .................................................. 163

Eruptive lichen planus in a child with celiac disease  
Dipankar De, Amrinder J. Kanwar ................................................................. 164

Xerosis and pityriasis alba-like changes associated with zonisamide  
Feroze Kaliyadan, Jayasree Manoj, S. Venkitakrishnan ................................ 165

Treatment of actinomycetoma with combination of rifampicin and co-trimoxazole  
Rajiv Joshi ...................................................................................................... 166

Author’s reply  

Vitiligo, psoriasis and imiquimod: Fitting all into the same pathway  
Bell Raj Eapen .................................................................................................. 169

Author’s reply  
Engin Şenel, Deniz Seçkin .............................................................................. 169

Multiple dermatofibromas on face treated with carbon dioxide laser: The importance of laser parameters  
Kabir Sardana, Vijay K. Garg ........................................................................ 170

Author’s reply  

Alopecia areata progressing to totalis/universalis in non-insulin dependent diabetes mellitus (type II): Failure of dexamethasone-cyclophosphamide pulse therapy  
Virendra N. Sehgal, Sambit N. Bhattacharya, Sonal Sharma, Govind Srivastava, Ashok K. Aggarwal .............................................................. 171

Subungual exostosis  
Kamal Aggarwal, Sanjeev Gupta, Vijay Kumar Jain, Amit Mital, Sunita Gupta ................................................................. 173
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Clinicohistopathological correlation of leprosy
Amrish N. Pandya, Hemali J. Tailor ................................................................. 174

RESIDENT’S PAGE

Dermatographism
Dipti Bhute, Bhavana Doshi, Sushil Pande, Sunanda Mahajan, Vidya Kharkar ................................................................. 177

FOCUS

Mycophenolate mofetil
Amar Surjushe, D. G. Saple ......................................................................................... 180

QUIZ

Multiple papules on the vulva
G. Raghu Rama Rao, R. Radha Rani, A. Amareswar, P. V. Krishnam Raju, P. Raja Kumari, Y. Hari Kishan Kumar .............................................................................................................. 185

EIJVDL

Net Study
Oral isotretinoin is as effective as a combination of oral isotretinoin and topical anti-acne agents in nodulocystic acne
Rajeev Dhir, Neetu P. Gehi, Reetu Agarwal, Yuvraj E. More .............................................................................................................. 187

Net Case
Cutaneous diphtheria masquerading as a sexually transmitted disease
T. P. Vetrichevvel, Gajanan A. Pise, Kishan Kumar Agrawal, Devinder Mohan Thappa ......................................................................................................................... 187

Net Letters
Patch test in Behcet’s disease
Ülker Gül, Müzeyyen Gönül, Seray Külçü Çakmak, Arzu Kılıç .............................................................................................................. 187

Cerebriform elephantiasis of the vulva following tuberculous lymphadenitis
Surajit Nayak, Basanti Acharjya, Basanti Devi, Satyadarshi Pattnaik, Manoj Kumar Patra ......................................................................................................................... 188

Net Quiz
Vesicles on the tongue
Saurabh Agarwal, Krishna Gopal, Binay Kumar .............................................................................................................. 188
Case Report

Non-familial multiple keratoacanthomas in a 70 year-old long-term non-progressor HIV-seropositive man

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ABSTRACT

We describe here multiple keratoacanthomas in an Human Immunodeficiency Virus (HIV)-seropositive 70 year-old man. The patient had multiple epithelial tumors of the skin showing rapid growth, histopathological features of a keratoacanthoma and a conspicuous tendency toward spontaneous remission. A diagnosis of nonfamilial multiple keratoacanthoma was established. The patient had a CD4 count of 633 cells/µL. The HIV disease in our patient was of a nonprogressive nature with CCR5-positive T cells.

Key Words: HIV infection, Multiple keratoacanthoma, Non-familial type

INTRODUCTION

Keratoacanthoma is an epithelial tumor of the skin characterized by rapid growth, histopathology similar to that of squamous cell carcinoma and a tendency towards spontaneous remission.[1] Keratoacanthomas have been noted in patients with immunosuppression but rarely in the HIV-infected.[2] We report here a case of non-familial multiple keratoacanthomas in a 70 year-old, long-term non-progressor, HIV-seropositive male patient.

CASE REPORT

A 70 year-old male presented with a few pinhead-sized, itchy, papular lesions on his left wrist and the adjoining forearm. Within a week, similar lesions appeared elsewhere. These lesions increased in size over three weeks to form nodules of variable sizes (1-1.5 cm). Sexual history of the patient revealed exposure to multiple extramarital sexual contacts with female sex workers while working in Mumbai from 1958 to 1968 at his age of 28 to 38 years. There was no history of any blood transfusion, surgery, fever, diarrhea, loss of weight, joint pains or any other systemic disease including diabetes. The patient was a chain smoker for the last 25 years. He denied any history of skin diseases or sexually transmitted infections in the past. There was no history of similar lesions observed in his parents or siblings. The patient’s HIV status was noticed two years ago only after the death of his wife (HIV status unknown). The patient’s children had died young at ages below five years; hence, no information was available about their HIV status.

On examination, the patient looked healthy for his age and had no pallor. There was clubbing of the fingernails. He had bilateral 2-3 cm-in size, soft, nontender, mobile, discrete inguinal lymph nodes. Cutaneous examination revealed multiple skin-colored and a few pigmented papules and nodules of various sizes on the dorsal aspect of both hands, the extensor aspect of the left forearm, the medial side of both legs, on the left foot and upper back [Figures 1 and 2]. Some of the lesions on the left leg had central crusting while the lesions on the forearm, both legs and the back showed umbilication. The papules and nodules on the left leg showed grouping and Köbner’s phenomenon [Figure 1].

Investigations revealed normal complete hemogram, liver and kidney function tests and blood sugar levels; VDRL was nonreactive. Anti-HIV-1 antibodies were detected by using both ELISA and Western Blot. Rapid test for CCR5 cell surface
marker was positive in lymphocytes using flow cytometry (conducted at the National Institute of Immunology, New Delhi). The CD4 count was 633 cells/µL, CD8: 1568 cells µL, CD3: 2271 cells/µL, CD4:CD8 = 0.40. Mantoux test was positive (13 mm × 14 mm) and sputum for acid-fast bacilli tested negative on three occasions. The X-ray chest (PA view), ultrasound (abdomen and pelvis) and sigmoidoscopy were normal.

The histopathological evaluation from one of the nodules showed an acanthotic epidermis with irregular elongations of the rete ridges. There was irregular epithelial proliferation in the islands of stratified squamous epithelium with glassy cytoplasm [Figures 3-4]. Individual cell keratinization was also observed. The dermis showed the presence of a chronic inflammatory infiltrate. There was no human papilloma virus (HPV) DNA in the skin tissue samples analyzed by means of the polymerase chain reaction (PCR) technique.

During the patient’s stay in the wards, there were fresh crops of similar lesions followed by resolution. However, some of the bigger lesions were treated with cryotherapy using liquid nitrogen for quicker resolution. In the next two months’ follow-up period after discharge from the hospital, there was almost complete resolution of all lesions.

**DISCUSSION**

Keratoacanthoma commonly occurs in the elderly as a solitary lesion; however, occasionally, there are several lesions growing rapidly within a few weeks. There are two variants of multiple keratoacanthoma: the self-healing epitheliomas (Ferguson-Smith) type and the eruptive Grzybowski type. Both variants are rare. The Ferguson-Smith type is a familial form, which affects both sexes with equal severity. Most cases develop during childhood and
adolescence and are characterized by the appearance of multiple, sometimes hundreds of keratoacanthomas. This disorder is inherited in an autosomal dominant form and has been described only in a few Scottish families. The eruptive Grzybowski type is characterized by tiny (2-3 mm diameter) and numerous follicular keratotic papules disseminated all over the body including the oral mucosa. In our case, the lesions clinically and histopathologically resembled solitary keratoacanthoma although they were multiple. In our patient, lesions of all three stages were observed during his continuous four months’ stay indoors.

Graells et al. reported a case of Muir-Torré syndrome in an HIV-positive male, who presented with a solitary keratoacanthoma and multiple sebaceous adenomas without any visceral neoplasm. Our patient with multiple keratoacanthomas was evaluated for the presence of the typical traits of Muir-Torré syndrome but there was no cutaneous sebaceous tumor or any other carcinoma of the GI tract, lung and genitourinary tract.

Keratoacanthomas have also been reported in immunosuppressed patients after bone marrow transplantation cyclosporine treatment or infection with HIV, thus suggesting that immunosuppression may play an etiologic role in some cases. Payne et al. reported two homosexual HIV-positive men with solitary keratoacanthoma, one preceded an epidermal cyst on the cheek while the chest was affected in the other patient. Studies by Payne et al. and others showed an association of keratoacanthoma with human papilloma virus (HPV) 25, HPV-19 and HPV-48 in lesions arising in HIV-infected patients. The use of PCR, cutaneous HPV DNA was detected in only 51% (37/72) of the keratoacanthomas. In our case, the isolation and typing of HPV by PCR revealed a negative result, which was in corroboration with study by Lus et al. who did not find any association between known HPV types and keratoacanthoma.

The atypical features in our case were multiple keratoacanthomas occurring in crops for a few months and the prolonged asymptomatic phase of the HIV disease itself. The patient possibly acquired HIV infection thirty-five years ago and was still asymptomatic and disease-free but for the continuous four months’ stay indoors.

It is difficult to ascertain the cause and association of HIV and multiple keratoacanthoma in the absence of HPV infection, significant immunosuppression due to HIV infection (nonprogressive) and Muir-Torre syndrome as in this case. It is possible that HIV infection could have influenced the appearance of keratoacanthomas over several months. To the best of our knowledge, this is the first case of an association of HIV infection with multiple keratoacanthomas.

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