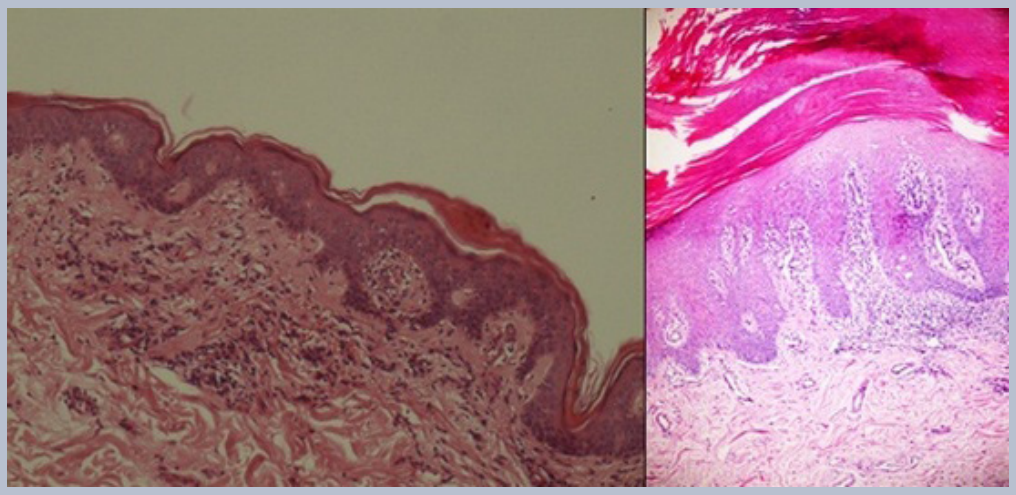
Volume 6, Number 4, September 2015
p. 384 - 497
Issue online since Mondday, September 28, 2015

ISSN: 2081-9390
DOI: 10.7241/ourd
Universal Impact Factor {UIF} (= 0.7319)

Our Dermatology Online

www.odermatol.com



4 / 2015

Our Dermatology Online

Editorial Pages

e-ISSN: 2081-9390
DOI: 10.7241/ourd

Quarterly
Our Dermatol Online

published since 01/06/2010 years

www.odermatol.com

Indexed in:

Universal Impact Factor for year 2012 is = 0.7319
system of opinion of scientific periodicals INDEX COPERNICUS (6,51)
(Academic Search) EBSCO
(Academic Search Premier) EBSCO
MNiSW (kbn)-Ministerstwo Nauki i Szkolnictwa Wyższego (4.00)
DOAJ (Directory of Open Acces Journals)
Geneva Foundation for Medical Education and Research (GFMER), Google Scholar, Open J-Gate, NewJour,
International Committee of Medical Journal Editors (ICMJE), Genamics JournalSeek, Hinari,
Bielefeld Academic Search Engine (BASE), WorldCat, e -journal, WorldWideScience.org, National Science Library,
LibSearch, Sciencegate, Virtual Science Library (VSL), Wanfang Data, COnnecting REpositories (CORE),
CAB Abstracts, Global Health, Journal Indexed in Directory of Research Journals Indexing,
OAIster: The Open Access Initiative, OAJSE - Open Access Journals Search Engine, Scirus

Previous website:	issue 1.2010	www.ndermatol.like.pl
	since issue 2.2010 to issue 3.2011	www.odermatol.like.pl
	since issue 4.2011	www.odermatol.com
Previous shortcut:	since issue 1.2010 to issue 3.2011	N Dermatol Online
	since issue 4.2011	Our Dermatol Online

Editorial Board

Contents

Contents

Our Dermatology Online

Original Article

A study of Onychomycosis in Krishna district of Andhra Pradesh, India

Dusi Ratna Harika[1], Anaparthy Usharani[2]

[1]Asst.Professor, Department of Microbiology, Dr. Pinnamaneni Siddhartha Institute of Medical Sciences & Research Foundation, Chinnaoutapalli, Krishna district, India, [2]Department of Microbiology, Rangaraya Medical College, Kakinada, India

Corresponding author: Prof. Anaparthy Usharani MD, E-mail: usharani.anaparthy@gmail.com

ABSTRACT

Introduction: Onychomycosis is a chronic infection of nails caused by fungi such as dermatophytes, yeasts or non-dermatophyte moulds. It is the most prevalent of all the nail ailments and affects 3- 5% of the population worldwide and represents 20-40% of onychopathies and about 30% of mycotic cutaneous infections. **Aims:** 1)To isolate and identify the etiological fungi and to assess the prevalence of onychomycosis. 2)To analyse the epidemiological and mycological features of onychomychosis. **Methods:** 109 nail samples were collected from 102 clinically suspected cases of onychomycosis and further analysed. **Results:** Of 102 cases, the commonest age group was 41-50 years 38 (37.25%); males were 63 (61.76%) and females 39 (38.24%); involvement of toe nails in 73 (71.57%), finger nails 25 (24.51%) and both 4 (3.92%); 56 (54.90%) belonged to low socio-economic status, middle 31 (30.39%) and high 15 (14.71%). Labourers were 14 (13.73%), farmers and office personnel 10 (9.80%). Of 109 samples, direct microscopy by KOH mount was positive in 82 (75.23%) and fungal culture in 52 (47.71%) of which 29 (26.61%) yielded dermatophytes, NDM's 11(21.15%), *Candida spp.* 8 (15.38%) and mixed growth 4 (7.68%). Dermatophytes 25(48.08%) were the predominant group isolated from toe nails and *Candida spp.* 6 (11.54%) from the finger nails respectively. Among the 56 isolates, dermatophytes were the predominant group 31 (55.36%) followed by NDM's 15 (26.78%) and *Candida* spp.10 (17.86%). **Conclusion:** Onychomycosis is a frequent cause of nail infection. The mycological study and the identification of etiological agents of onychomycosis are needed to confirm the clinical diagnosis and for the choice of therapy.

Key words: Onychomychosis; Dermatophytes; Candida spp; Non dermatophyte molds

INTRODUCTION

Fungi are ubiquitous in nature. The term 'Onychomycosis' is derived from Greek word 'onyx', nail and 'mykes', fungus [1]. Onychomycosis comprises all fungal infections affecting the nail apparatus, i.e., nail matrix, nail plate, cuticle, mesenchymal tissue & nail folds [2] and can be caused by dermatophytes, yeasts or non-dermatophyte moulds.

Onychomycosis has been referred to as the most prevalent of all the nail ailments and affects 3- 5% of the population worldwide [3] and represents 20-40% of onychopathies and about 30% of mycotic cutaneous infections [4]. Various workers have reported the incidence to vary from 0.5 to 5% in the general population in India [5]. The prevalence rate of onychomycosis is determined by age, predisposing factors, social class, occupation, climate, living environment and frequency of travel. Several factors have been implicated to the increase in disease such as reduced peripheral circulation, diabetes, nail trauma and difficulty to maintain proper nail hygiene [6]. Nail diseases can lead to impairment of hand function, difficulty in walking, and cosmetic disfigurement.

Dermatophytes are the most frequently implicated causative agents in onychomycosis approximately 90% in toe nail and 50% in finger nail infections [7]. The hyphae of the dermatophytes penetrate the stratum corneum of the skin and nails. The families

How to cite this article: Ratna Harika D, Usharani A. A study of onychomycosis in Krishna district of Andhra Pradesh, India. Our Dermatol Online. 2015;6(4):384-391.
Submission: 01.04.2015; **Acceptance:** 20.06.2015
DOI: 10.7241/ourd.20154.105

that include many of the known keratinolytic fungi are the Arthrodermataceae and Onygenaceae in the phylum Ascomycota.Members of these families are homogenous with respect to appearance, physiology, taxonomy, antigenicity, basic growth requirements, infectivity, and the diseases they cause[8]. Being a pathogen and a colonizer, *Candida albicans* is also found in the environment, particularly on leaves, flowers, water, soil and infect more fingernails than toenails. Moulds are distributed worldwide. While they are commonly considered contaminants, they also cause infections in immunocompromised patients and in the elderly with damaged nail integrity [9]. The disease is more frequent among men than women & it increases with age [10]. Toe nails are involved in the majority of cases. The ratio of finger nail to toe nail onychomycosis was found to be 1:4 [1].

Superficial fungal infections of the nail affect millions of individuals worldwide. In onychomycosis, infected nails serve as a chronic reservoir of infection which can give rise to repeated mycotic infections of the skin. It is of significance to suspect onychomycosis, perform mycological diagnosis and undertake treatment. This may help to prevent nail dystrophy and the spread of infection. Though onychomycosis is rarely life threatening, its chronicity, resulting cosmetic disfigurement and morbidity makes it an important public health problem.

MATERIALS AND METHODS

Ethical Consideration

The study was reviewed and approved by the Institutional Ethical Committee, Siddhartha Medical College and Government General Hospital, Vijayawada, Krishna district.

Inclusion Criteria

i) Clinically diagnosed cases of onychomycosis having destruction of nail plate, onycholysis, subungual hyperkeratosis, discoloration & thickening of nail plate alone or in combination.ii) Patients of both sex and all ages. iii) Patients who were not on antifungal therapy.

Exclusion Criteria

i) Patients who had received treatment either with topical and/or systemic antifungal agents for present nail condition within the last one month. ii) Diagnosed cases of other dermatological diseases having nail changes eg. Psoriasis, Lichen Planus, Eczema,etc.

All cases with onychomycosis attending in Department of Dermatology, Government General Hospital, Vijayawada, Krishna dist., Andhra Pradesh from September 2011 to August 2012 were examined and selected based on history, clinical examination, inclusion and exclusion criteria. One hundred and nine samples from one hundred and two clinically suspected cases of onychomycosis (Fig. 1) were included in the present study. Nail clippings or subungual scrapings from all these cases were collected with a surgical blade after cleaning the affected area with 70%alcohol from the involved nail bed and from the undersurface of the nail.

The specimens were processed by direct microscopic examination using 20% KOH (Fig. 2) and isolation by culture. Each of the samples was inoculated into two slopes of modified Sabouraud's dextrose agar (SDA, Himedia laboratories) slants, one with gentamycin and another with gentamycin and cycloheximide. Cultures were incubated at 25°C and 37°C and examined daily for first week and twice a week for 6wks.

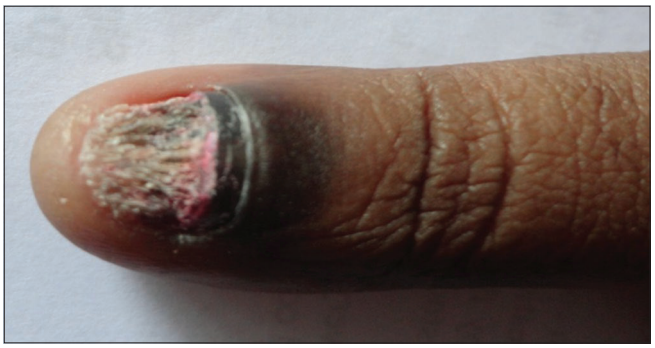


Figure 1: Onychomycosis affecting finger nail.

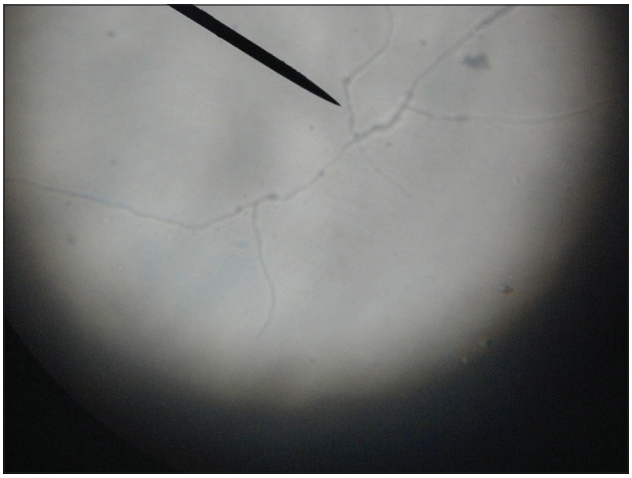


Figure 2: Direct – KOH wet mount, (40X).

Each of the SDA tube was observed for texture, colony morphology, obverse and reverse pigmentation. If growth was present, a LPCB teased mount was prepared and examined under microscope. Gram stain was done and germ tube test performed when the growth was creamy. Yeast-like growths of the isolates were examined by Gram staining (Fig. 3), inoculated onto Hichrome agar (Fig. 4), cornmeal agar and SDA broth for presumptive identification of Candida species and their differentiation respectively. Slideculture was put up for mycelial isolates to study the undisturbed morphological details of the fungi. Biochemical reaction such as Urease test was done for differentiation of dermatophytes. Repeated cultures were done for confirmation of non-dermatophytic moulds (Figs 5 - 8).

Identification

Growth on SDA with actidione and antibiotics were identified as dermatophytes (Fig-9) as etiologic agents, after observing microconidia and macroconidia under LPCB mount. The identification was confirmed by micromorphological aspects on slide culture and urease test which is positive with *T.mentagrophytes* and negative with *T.rubrum*. *Candida* species were identified by observation of budding yeast cells and pseudomycelium under light microscopy with KOH, yeast-like growth on SDA medium and by gram positive budding yeast cells on Gram stained smear of the culture. Growth in the cyclo-heximide free medium indicates that the infective agent may be an NDM. The identification of non-dermatophytic fungi were confirmed by following micro and macroscopic evaluations of the primary cultures and slide culture. When the light microscopy of a nail specimen showed filaments with only a non-dermatophytic growth in culture, a second nail specimen was examined again by light microscopy and culture to confirm non-dermatophytic mould infection.

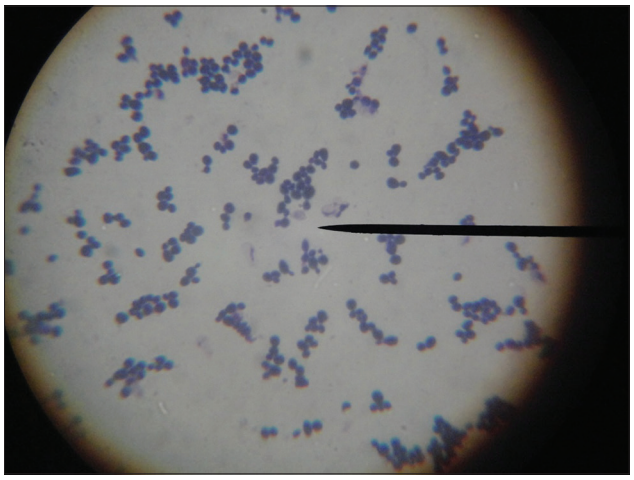


Figure 3: Gram positive budding yeast cells on Gram's staining.

Figure 4: Growth of different *Candida spp.* on Hichrome agar.

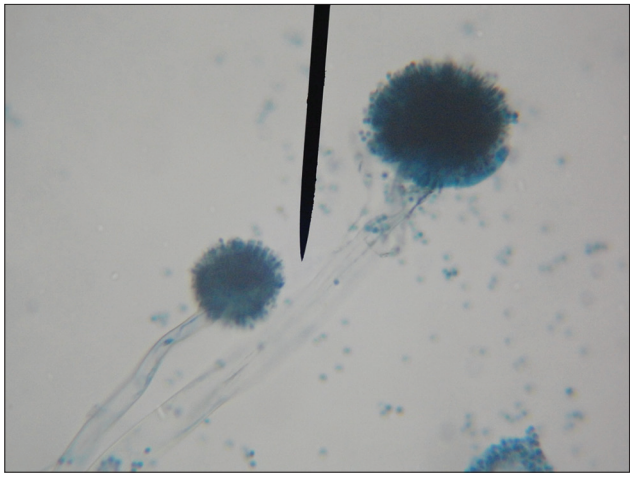


Figure 5: *Aspergillus niger* LPCB, (40X).

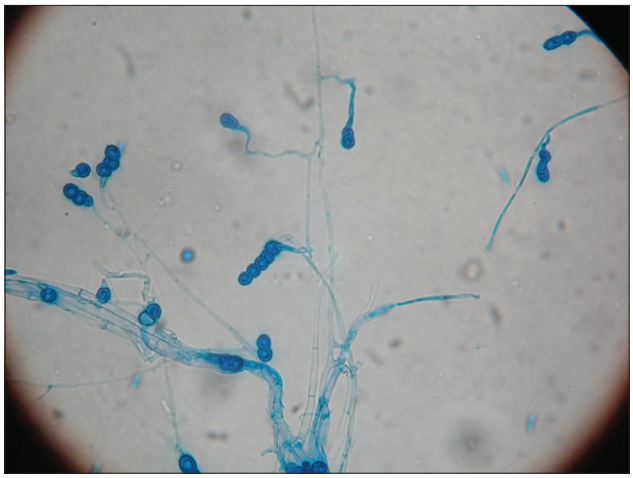


Figure 6: *Scopulariopsis brevicaulis* undisturbed morphology LPCB, (40X).

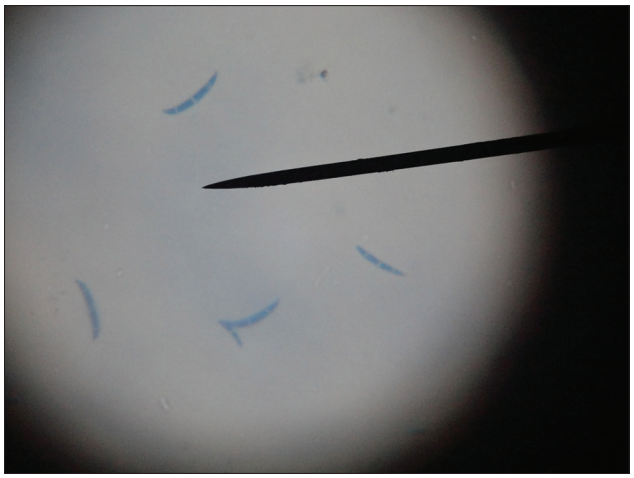


Figure 7: *Fusarium spp.* LPCB, (40X).

Figure 8: *Curvularia spp.* LPCB, (40X).

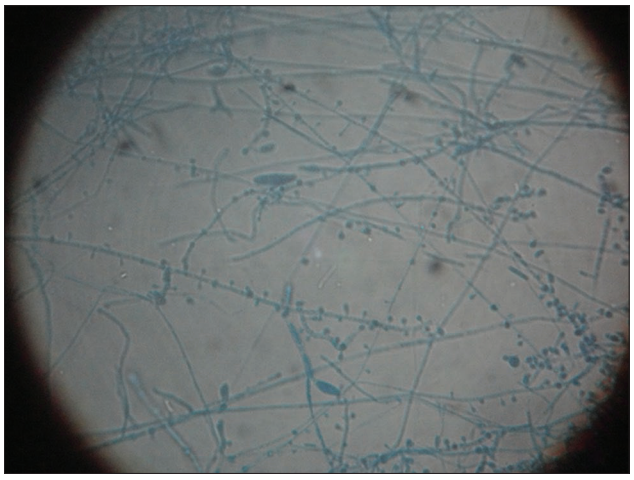


Figure 9: *Trichophyton mentagrophytes* LPCB, (40X).

Ethics

This study was performed on human subjects; thus, all patients were aware of the presence of the study and they were fully informed about the drug and its side-effects.

RESULTS

Of 102 cases (Table I), 41-50 years 38 (37.25%) were the commonest age group followed by 51-60 years 26 (25.49%) with males 63 (61.76%) and females 39 (38.24%) and the ratio of male to female onychomycosis patients was approximately 1.6:1. It was more common in people of low-socio economic status 56(54.90%) followed by middle 31 (30.39%) and high 15 (14.71%).

Of the 102 cases, toe nails were the most frequent anatomic site involved in 73 (71.57%) cases followed by finger nails in 25 (24.51%) cases and both 4 (3.92%) cases.

Of the 102 infected cases of onychomycosis, 109 samples (80toe nails,29 finger nails) were collected of which 38 (37.26%) are associated with occupations due to increased physical activity followed by other occupations 37 (36.28%) and occupations associated with wet work 27 (26.47%) (Table II).

Distal and lateral subungual onychomycosis (DLSO) was the commonest clinical type 88 (80.73%) and was followed by Paronychia 9 (8.26%), Total dystrophic onychomycosis (TDO) 7 (6.42%), Proximal subungual onychomycosis (PSO) 3 (2.75%) and Superficial white onychomycosis(SWO) 2(1.83%). Both DLSO & Paronychia were most commonly seen in toe nails accounting for 61.47% and 5.50% respectively (Table III).

Direct microscopy by KOH mount was positive in 82 (75.23%) cases and fungal culture in 52 (47.71%) cases (Table IV). From 52 culture positive samples, 56 fungal isolates were obtained (Table V). Dermatophytes 25 (48.08%) were the predominant group isolated from toe nails followed by non-dermatophyte moulds 8 (15.38%) and *Candida spp.*2 (3.85%). In the finger nails, *Candida spp.* 6 (11.54%) were the predominant group isolated followed by dermatophytes 4 (7.7%) and non-dermatophyte moulds 3(5.7%). Mixed growth of two different fungi was observed in a single case of finger nail infection 1 (1.92%) and 3 cases of toe nail infection of 1.92% each.

Among the 56 isolates (Table VI), dermatophytes were the predominant group 31 (55.36%) with *T. rubrum* the most common species isolated 19(33.93%) followed by *T. mentagrophytes* 11 (19.64%) and *M. nanum* 1 (1.79%). Non-dermatophyte filamentous fungi

Table 1: Distribution of cases with onychomycosis in relation to age, gender, socio- economic status, nail involvement (n=102)

Study group	No	Percentage
Age in years		
11-20	1	0.98
21-30	12	11.77
31-40	22	21.57
41-50	38	37.26
51-60	26	25.49
61-70	3	2.94
Gender		
Males	63	61.76
Females	39	38.24
Socio economic		
Low	56	54.90
Middle	31	30.39
High	15	14.71
Toe nails	73	71.57
Finger nails	25	24.51
Both	4	3.92

Table 2: Distribution of cases according to occupation (n=102)

Category	No. of cases (%)
1) Associated with increased physical activity	
Manual labourers	14 (13.73)
Farmers	10 (9.80)
Mechanics	3 (2.94)
Vehicle operators	6 (5.88)
Tailors	5 (4.90)
Total	38 (37.26)
2) Associated with wet work	
House wives	7 (6.86)
Domestic helpers	7 (6.86)
Washer men	4 (3.92)
Cooks	2 (1.96)
Hotel workers	4 (3.92)
Gardeners	3 (2.94)
Total	27 (26.47)
3) Others	
Office personnel	10 (9.80)
Businessmen	7 (6.86)
Students	3 (2.94)
Professionals	9 (8.82)
No specific occupation	8 (7.84)
Total	37 (36.28)
Total cases	102

Table 3: Clinical patterns of onychomycosis (n=109)

Patterns	Toe nails (n=80) (%)	Finger nails (n=29) (%)	Total (%)
DLSO	67 (61.47)	21 (19.27)	88 (80.73)
PSO	1 (0.92)	2 (1.83)	3 (2.75)
SWO	2 (1.83)	0	2 (1.83)
TDO	4 (3.67)	3 (2.75)	7 (6.42)
Paronychia	6 (5.50)	3 (2.75)	9 (8.26)

DLSO: Distal lateral subungual onychomycosis, PSO: Proximal subungual onychomycosis, SWO: Superficial white onychomycosis, TDO: Total dystrophic onychomycosis

15 (26.78%) constitute the second predominant group with *Curvularia spp.*3 (5.36%), *Aspergillus niger, Bipolaris spp.* and *Scopulariopsis brevicaulis* of 2 (3.57%) each and occasionally *Aspergillus terreus, A.flavus, Alternaria spp., Fusarium spp., Scytalidium spp.* and *Scedosporium apiospermum* of 1 (1.79%) each were isolated. The other group of fungi isolated were the *Candida spp.* 10 (17.86%) with *C.albicans* 6 (10.71%),*C.krusei* 2 (3.57%), *C.tropicalis* and *C.glabrata* of 1 (1.79%) each.

Table 4: Microscopy and Culture positivity of the clinical samples (n=109)

Direct examination	Fungal culture (%)		Total (%)
	Positive	Negative	
KOH positive	43 (39.45)	39 (35.78)	82 (75.23)
KOH negative	9 (8.26)	18 (16.51)	27 (24.77)
Total	52 (47.71)	57 (52.29)	109

DISCUSSION

Onychomycosis is a chronic infection of the nails; nowadays considered a serious problem for public health, in view of its high occurrence in the worldwide population. The prevalence is probably higher than currently thought, as the difficulty in clinical-mycological diagnosis, inappropriate collection of material for analysis as well as ineffective treatment make it hard to ascertain the true profile of such onychopathies.

This study was a prospective study conducted in the Department of Microbiology on one hundred and two cases of clinically diagnosed/suspected onychomycosis in an attempt to study the epidemiology of this disorder in the general population in and around Vijayawada, Andhra Pradesh.

In the present study of 102 cases (Table I), onychomycosis was common in the age group of 41-50 years 38 (37.26%) followed by 51-60 years 26 (25.49%). Although many reports indicate that the prevalence of onychomycosis increases with age, with the highest prevalence among the elderly more than 60 years old [7], there was a decreasing prevalence in patients over the age of 50 years in the present study. The increase in cases with age may be justified by the presence of antecedent diseases like DM, peripheral vascular disease, personal habits such as chronic smoking, trauma to the aged nails.

Of the 102 cases (Table I), there were more men 63 (61.76%) than women 39 (38.24%). Similar finding have been reported by some authors [2,10] with a

Table 5: Onychomycosis due to different fungal groups: number of samples and isolates

Nails	Dermatophytes (DM) (%)	Non-dermatophyte molds (NDM) (%)	*Candida* spp (C) (%)	Mixed (%)				Total culture positive samples (%)	Total isolates
				DM+C	DM+NDM	NDM+NDM	NDM+C		
Finger nails	4 (7.7)	3 (5.7)	6 (11.54)	1 (1.92) (2 isolates)	0	0	0	14 (26.92)	15
Toe nails	25 (48.08)	8 (15.38)	2 (3.85)	0	1 (1.92) (2 isolates)	1 (1.92) (2 isolates)	1 (1.92) (2 isolates)	38 (73.08)	41
Total	29 (55.77)	11 (21.15)	8 (15.38)	1 (1.92)	1 (1.92)	1 (1.92)	1 (1.92)	52 (100)	56

Table 6: Fungal isolates obtained from the clinical samples (n=56)

Dermatophytes	No.of isolates (%)	*Candida spp.*	No.of isolates (%)
T.rubrum	19 (33.93)	*C. albicans*	6 (10.71)
T.mentagrophytes	11 (19.64)	*C. krusei*	2 (3.57)
M.nanum	1 (1.79)	*C. tropicalis*	1 (1.79)
		C. glabrata	1 (1.79)
Total	31 (55.36)	Total	10 (17.86)
Non- dermatophyte moulds (NDM)			
Alternaria spp.	1 (1.79)	*Curvularia spp.*	3 (5.36)
A. niger	2 (3.57)	*Fusarium spp.*	1 (1.79)
A. terreus	1 (1.79)	*S. brevicaulis*	2 (3.57)
A. flavus	1 (1.79)	*Scytalidium spp.*	1 (1.79)
Bipolaris spp.	2 (3.57)	*S. apiospermum*	1 (1.79)
Total			15 (26.78)
Total isolates			56

prevalence of 75.4% and 65% respectively among men. However, few others [4,11] have reported a high prevalence among women (62.7% and 51.96%). The ratio of male to female onychomycosis in the present study was approximately 1.6:1 which coincides well with a study [12] of 1.63:1. Higher incidence in males may be because they are more exposed to outdoors with greater physical activity and are more prone to trauma of nails.

Onychomycosis was more common in people of low socioeconomic status 56(54.90%) in the present study (Table I) when compared to middle 31 (30.39%) and high 15 (14.71%), the reason being the higher prevalence of poor hygienic practices and overcrowding in this study group. Toe nails are about seven times more frequently involved than finger nails due to three times slower growth rate [13]. In the present study, toe nails were the most frequent anatomic site involved in 73 (71.57%) cases and finger nails in 25 (24.51%) cases and both nails concurrently in 4 (3.92%) cases (Table I).

Manual labourers 14 (13.73%) were the predominant group followed by farmers and office personnel 10 (9.80%) each in the present study (Table II). Labourers on the other hand have increased perspiration, a greater risk of occupation- related trauma and exposure to soil saprophytes; while the use of occlusive footwear by office personnel might predispose them to onychomycosis. And in a similar study from Visakhapatnam [11], there was a higher prevalence in house wives(33.33%) as household wet work also appears to be an important predisposing factor.

In the present study,109 samples (80toe nails, 29 finger nails) were collected from 102 cases as some of the cases had involvement of multiple sites and culture was done consequently. As per Table-III, the most frequent clinical presentation was DLSO 88 (80.73%) followed by Paronychia in 9 (8.26%)cases, a finding which is in consonance with earlier reports [2] from Himachal Pradesh. In contrast, DLSO (50%) was the commonest presentation followed by PSO (20.4%) in a study [10] from Aurangabad.

The two conventional methods used for identification of fungi were direct microscopy with KOH mount and fungal culture.Our study revealed a mycological positivity of 75.23% on direct microscopic examination(Table-IV) which was very sensitive for showing the presence of fungi. But it was lower than the results of various researchers [10,14] who reported 81.8% &82.35% respectively, while this may be considered high when compared with the study [6] showing mycological positivity of 34%. Culture positivity in the present study was 47.71%, including 43(39.45%) with positive direct examination and 9 (8.26%) with negative direct examination as per Table IV which nearly correlated with the study [10] showing 48.8%. In contrast to this, some authors [12,15] have reported a comparatively high culture positivity rate of 62.7% and 60% respectively.

In the present study, as per Table-V has documented that dermatophytes 25 (48.08%) were the predominant group isolated from toe nails and *Candida spp.* 6 (11.54%) from the finger nails respectively, a similar finding in common with reports [4,6] from Brazil and Delhi. The lesions due to dermatophytes commonly occur on feet as the warmth, moisture promote the contamination; sweating, cramped or tight fitting foot ware or rubber

shoes prevent sweat evaporation and create the ideal environment for dermatophytoses. Local factors such as repeated damage of cuticle due to manicure, contact with substances containing sugars, wet work like washing, cooking, etc., and hyperhidrosis promote the infection with *Candida* [16].

As with dermatophyte infections, mould infections are much more common in the toe nails 8 (15.38%) than in the finger nails 3 (5.7%) in the present study (Table V). Non-dermatophyte moulds are filamentous fungi that are commonly found in nature as soil saprophytes and plant pathogens. Because these moulds are not keratinolytic, unlike dermatophytes, they only live on unkeratinized intercellular cement or must take advantage of previous keratin destruction by dermatophytes, trauma, or another nail disease i.e. these fungi act as secondary pathogen. Therefore, the isolates were confirmed after two repeated cultures of two different samples from the cases at different intervals. Reports from Tehran [17] and Turkey [18] show that mould onychomycosis is more frequent in toe nails than in finger nails and in the elderly. This situation can be prone due to more traumas to the nail with age and foot wear and also in labourers, and corresponds to very slow growth of finger and toe nail plates as well as to much higher incidence of impaired blood supply to the extremities.

In 7.68% of cultures, a mixture of dermatophytes or yeast-like or moulds were isolated. A single case of mixed infection of *T.rubrum* and *C.albicans* was observed in a finger nail. And the mixed isolates in the toe nails were geophilic *M.nanum* and *Scedosporium apiospermum* accounting for 1.79% of cultures. *Fusarium* and *Scopulariopsis brevicaulis* of 1.79% & *C.albicans* and *Curvularia spp.* of 1.79% (Table V).Various authors from Turkey [18] and Indonesia [19] have reported mixed infections of dermatophytes and *Candida spp.* as 2% and 2.21% of cultures respectively and 3 cases(5.55%) of mixed infections of *T.rubrum* and *Aspergillus niger* have been reported from Jammu [20], all of which were low when compared with the present study. The most probable explanation for mixed etiology is that the diseased and dystrophic nails already damaged by dermatophytes are easily invaded by non-dermatophytic moulds.

Among the 56 isolates in the present study as per Table VI, dermatophytes were the predominant isolates 31 (55.36%) followed by non-dermatophyte moulds 15 (26.78%) and *Candida spp.* 10 (17.86%). A high incidence of onychomycosis due to dermatophytes was reported in studies from Delhi & Sikkim [6,14] while *Candida spp.* were predominant in a study from Visakhapatnam [11] as this can be attributed to the fact that epidemiology of onychomycosis varies from one geographical region to other. The most common isolate obtained in the present study was *T.rubrum* 19 (33.93%) and it was followed by *T.mentagrophytes* 11(19.64%), a finding in accordance with reports [10] from Aurangabad with isolation rates of *T.rubrum* (34.88%) and *T.mentagrophytes* (25.58%).

Non-dermatophytic filamentous fungi constitute the second predominant group with an isolation rate of 26.78% and the organisms in decreasing order of frequency are *Curvularia* 3 (5.36%), *Aspergillus niger, Bipolaris spp.* and *Scopulariopsis brevicaulis* of 2 (3.57%) each, *A.terreus, A.flavus, Alternaria spp., Fusarium spp., Scytalidium spp. & Scedosporium apiospermum* of 1 (1.79%) each (Table VI). Studies from different parts of India showed isolation rates of different moulds as 27.91% from Aurangabad [10], 35.14% from Banglore [12], 35.71% from Sikkim [14] all of which were higher than the present study and this variation may reflect geographic differences in mould distribution. Suggested predisposing factors for NDM involvement include increasing age, poor peripheral circulation, immunosuppression, peripheral neuropathy and trauma [7].

The other group of fungi isolated were the *Candida spp.* 10 (17.86%) with *C.albicans* 6 (10.71%) as the predominant species followed by *C.krusei* 2 (3.57%), *C. tropicalis& C.glabrata* 1 (1.79%) one each in the present study (Table VI). It is believed that in tropical countries like India, hot humid climate may be a factor responsible for the causative role of yeasts.

The clinical diagnosis of onychomycosis should always be confirmed by direct microscopy and culture. Fungal culture of the subungual keratinous material provides final confirmation of the active cases of onychomycosis and enables to accurately pinpoint the causative organism. Compared to other studies, the low incidence of onychomycosis as seen in the present study may be because of low reporting, possibly due to less awareness and non hindrance of the problem to their occupation.

CONCLUSION

Onychomycosis is a frequent cause of nail infection. It is more prevalent in places where humidity and

warmth are present. The mycological study and the identification of etiological agents of onychomycosis are needed to confirm the clinical diagnosis and for the choice of therapy. The etiological spectrum of onychomycosis is largely dependent on the flora in the immediate environment of the individual. It is influenced by geographic, climatic and occupational factors. It is desirable to determine the prevalence of etiologic agents of onychomycosis in any particular area in order to develop adequate control measures.

Statement of Human and Animal Rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of Informed Consent

Informed consent was obtained from all patients for being included in the study.

REFERENCES

1. Chander J. Textbook of Medical Mycology. (Chandigarh): Mehta publishers, p 122-142; 266-283; 508-516, 2009.
2. Gupta M, Sharma NL, Kanga AK, Mahajan VK, Tegta GR. Onychomycosis: Clinico- mycologic study of 130 patients from Himachal Pradesh, India. Indian J Dermatol Venereol Leprol. 2007;73:389-92.
3. Grover C, Khurana A. Onychomycosis: Newer insights in pathogenesis and diagnosis. Indian J Dermatol Venereol Leprol. 2012;78:263-70.
4. Lopes JO, Alves SH, Mari CRD, Oliveira LTO, Brum LM, Westphalen JB, et al. A ten-year survey of onychomycosis in the Central Region of the Rio Grande do Sul, Brazil. Rev Inst Med Trop S Paulo. 1999;41:147-9.
5. Kaur R, Kashyap B, Bhalla P. Onychomycosis- epidemiology, diagnosis and management. Indian J Med Microbiol. 2008;26:108-16.
6. Kaur R, Kashyap B, Bhalla P. A five-year survey of onychomycosis in New Delhi, India: Epidemiological and laboratory aspects. Indian J Dermatol. 2007;52:39-42.
7. Elewski BE. Onychomycosis: Pathogenesis, diagnosis and management. Clin Microbiol Rev. 1998;11:415-29.
8. Rippon JW. Medical mycology: the pathogenic fungi and the pathogenic actinomycetes. Philadelphia, Pa: The W.B. Saunders Co., p. 169-275, 1988.
9. Effendy I, Lecha M, Feuilhade de Chauvin M, Di Chiacchio N, Baran R. Epidemiology and clinical classification of onychomycosis. J European Acad Dermatol Venereol. 2005;19:8–12.
10. Veer P, Patwardhan NS, Damle AS. Study of onychomycosis: Prevailing fungi and pattern of infection. Indian J Med Microbiol. 2007;25:53-6.
11. Jesudanam TM, Rao GR, Lakshmi DJ, Kumari GR. Onycomycosis: A significant medical problem. Indian J Dermatol Venereol Leprol. 2002;68:326-9.
12. Grover S. Clinicomycological evaluation of onychomycosis at Bangalore and Jorhat. Indian J Dermatol Venerol Leprol. 2003**;**69:284-6.
13. Singal A, Khanna D. Onychomycosis: Diagnosis and management. Indian J Dermatol Venereol Leprol. 2011;77:659-72.
14. Adhikari L, Gupta AD, Pal R, Singh T. Clinico-etiologic correlates of onychomycosis in Sikkim. Indian J Pathol Microbiol. 2009;52:194-7.
15. Sujatha V, Grover S, Dash K, Singh G. A clinico - Mycological evaluation of onychomycosis. Indian J Dermatol Venereol Leprol. 2000;66:238-40.
16. Ramani R, Kumari GR, Shivananda PG. Onychomycosis in coastal Karnataka. Indian J.Med Microbiol. 1993;11:223-5.
17. Bassiri-Jahromi S, Khaksar AA. Nondermatophytic moulds as a causative agent of onychomycosis in Tehran. Indian J Dermatol. 2010;55:140-3.
18. Hilmioğlu-Polat S, Metin DY, Inci R, Dereli T, Kilinç I, Tümbay E. Non-dermatophytic molds as agents of onychomycosis in Izmir, Turkey - a prospective study. Mycopathologia. 2005;160:125–8.
19. Bramono K, Budimulja U. Epidemiology of onychomycosis in Indonesia: Data obtained from three individual studies. Jpn J Med Mycol. 2005;46:171-6.
20. Ahmad M, Gupta S, Gupte S. A clinico-mycological study of onychomycosis. Egyptian Dermatol Online J. 2010;6:1-9.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Original Article

Weekly methotrexate versus daily isotretinoin to treat moderate-to-severe chronic plaque psoriasis: A comparative study

C. Abhinav, Vikram K Mahajan, Karaninder S. Mehta, Pushpinder S. Chauhan, Mrinal Gupta, Ritu Rawat

Department of Dermatology, Venereology and Leprosy, Dr. R. P. Govt. Medical College, Kangra (Tanda)-176001 (Himachal Pradesh), India

Corresponding author: Dr. Vikram K Mahajan, E-mail: vkm1@rediffmail.com

ABSTRACT

Introduction: There is paucity of data on efficacy of isotretinoin versus methotrexate in psoriasis patients. **Aims:** We compared the efficacy and safety of once a week methotrexate and daily isotretinoin for the treatment of moderate-to-severe chronic plaque psoriasis. **Methods:** 43 (M: F 30:13) consenting patients having moderate-to-severe chronic plaque psoriasis were divided on alternate basis in Group-A for treatment with methotrexate (15mg/week) and Group-B for isotretinoin (30mg/d) therapy. The clinical response was assessed by the percentage reduction in the baseline PASI scores during next 12 weeks. **Results:** 14 patients in Group-A and 11 patients in Group-B completed the study. The mean percentage reduction in the PASI score was 79.78 ± 20.68 in the methotrexate group and 51.92 ± 23.83 in the isotoin group at the end of 12 weeks. Five patients in Group-A achieved >75% reduction in the PASI score, while only 3 patients in Group-B showed similar results at the end of 12 weeks signifying faster disease clearance with methotrexate. Isotretinoin-related serious adverse effects were fewer and did not warrant treatment discontinuation. **Conclusions:** Isotretinoin may be an option for alternative therapy in patients who cannot afford acetretin, are intolerant to methotrexate, have achieved its cumulative dose, or in rotational therapy.

Key words: Isotretinoin; Methotrexate; Psoriasis; Retinoids

INTRODUCTION

Chornic plaque psoriasis is a common, genetically determined, inflammatory and proliferative dermatological disorder with an estimated prevalence of 1.5-3.0% in Europe and 2.2-2.6% in US [1,2]. Immunologically mediated activation of T lymphocytes is central to the inflammation in the dermal microenvironment and epidermal hyper-proliferation occurs secondary to the inflammatory events that follows a Th1 type of immune response [3]. It affects both genders at any age and has chronic and unpredictable clinical course adversely affecting quality of life. However, despite a substantial progress toward elucidation of genetic and pathophysiological pathways involved in psoriasis, a safe affordable and effective/curative therapy remains desirable. Most topical medications (emollients, tars, anthralins, corticosteroids, retinoids (tazarotene), vitamin-D analogues (calcipotriol, calcitriol, tacalcitol, maxacalcitol), topical calcineurin inhibitors (tacrolimus, pimacrolimus), narrowband UVB phototherapy) or systemic therapies (methotrexate, retinoids, cyclosporine A, PUVA (psoralens+UVA), hydroxyurea, biologics), used alone or in combination with topical therapies, control the severity and extent of psoriasis. These therapies are associated with potentially severe toxicities, require extensive monitoring and are expensive [1,4]. The biologics (alefacept, infliximab, adalimumab, etanercept, efalizumab) offer better safety profile but cost and availability remain prohibitory. Despite concerns for hepatotoxicity, methotrexate

How to cite this article: Abhinav C, Mahajan VK, Mehta KS, Chauhan PS, Gupta M, Rawat M. Weekly methotrexate versus daily isotretinoin to treat moderate-to-severe chronic plaque psoriasis: a comparative study. Our Dermatol Online. 2015;6(4):392-398.

Submission: 01.05.2015; **Acceptance:** 28.06.2015

DOI: 10.7241/ourd.20154.106

(7.5mg to 30mg/week) remains standard systemic therapy for moderate to severe psoriasis unresponsive to topical therapy, in psoriatic arthritis and nails psoriasis due to affordability, high efficacy and fast results [1,5]. There has been a continuing search for a less toxic, but equally effective replacement for methotrexate once its cumulative dose (1.0-1.5 gm) is achieved [6]. A rapid and impressive response may be seen with retinoids in pustular psoriasis and erythrodermic psoriasis. Etretinate, the first retinoid, was withdrawn because of potential risk of teratogenicity due to its long half-life. Acitretin, the active metabolite of etretinate, has a shorter half-life and can be taken for long when not planning pregnancy for 3 years after its discontinuation [7]. However, high cost of acitretin precludes its routine use in most instances. Comparatively, isotretinoin is cheaper and has shorter half-life and contraception is required during therapy and 1 month thereafter [8,9]. Although considered more effective in pustular psoriasis (8-10), its efficacy in chronic plaque psoriasis remains under studied. Moy et al [10] found acitretin (0.75 mg/kg/d for 8 weeks) effective in 18/19 of chronic plaque psoriasis and also achieved complete to moderate response with isotretinoin (1.5 mg/kg/d) in their 4/10 patients with chronic plaque psoriasis involving 20-50% body surface area when given for the same period. However, there is overall paucity of data on efficacy of isotretinoin in patients having moderate to severe chronic plaque psoriasis. In this prospective study, we compared the efficacy and safety of once a week methotrexate and daily isotretinoin for the treatment of moderate to severe chronic plaque psoriasis.

MATERIAL AND METHODS

Forty three consenting patients of psoriasis presenting with moderate to severe psoriasis over a period of one year were enrolled after written/informed consent. PsoriasisArea-and-Severity Index (PASI) score of 6-12 or body surface area (BSA) involvement up to 10% was considered moderate psoriasis while PASI score >12 or BSA involvement >10% was considered severe psoriasis [11]. The study was approved by the Institutional Protocol Review Board and Institutional Ethics Committee (Rgn no. ECR/490/Inst/HP/2013). Patients <18 years of age, having BMI >30 or weighing <30 kg, women who were pregnant, lactating or unwilling for an effective regimen of contraception, patients with psoriatic arthritis, any systemic disease (hepatorenal or hematologic impairment, deranged lipid profile acute infection requiring antimicrobial therapy, uncontrolled hypertension or diabetes mellitus, neoplasias, psychiatric illness), or a long history of alcohol intake were excluded. A detailed history and clinical examination were recorded in all eligible patients. Laboratory investigations were performed in all patients, biweekly during the first month and every month during the next two months and included complete blood counts, fasting blood sugar, lipid profile, serum creatinine, serum urea, serum glutamine pyruvate transaminase (SGPT), serum glutamine oxalate transaminase (SGOT), alkaline phosphatase, and serum bilirubin (total and conjugated). Chest X-ray and serum electrolytes were performed at the beginning and at the end of the treatment in each patient.

Forty-three patients were eligible for the study. They were assigned alternately to Group-A for weekly dose of methotrexate (Methotrexate Group) or Group-B for daily dose of isotretinoin (Isotretinoin Group) with option to withdraw any time from the study. Patients already on any antipsoriasis treatment were given a wash off period of 2 and 4 weeks for topical and systemic therapy respectively. Photographic records were kept in all patients before and after the treatment.

Treatment Protocols

Group-A patients received once a week oral methotrexate 15 mg (given in two divided doses at an interval of 12 hours) and Group-B patients received isotretinoin 30 mg/d PO. Patients were evaluated every two weekly for first 4 weeks and once in 4weeks thereafter for the next 8weeks. Side effects known to be associated with methotrexateor isotretinoin, as well as other side effects that seem relevant to the treatment were evaluated on every visit. The treatment was stopped immediately in the event of intolerable adverse drug reaction. Only emollients and antihistamines in both the groups and folate supplements and anti-emetics for Group-A (methotrexate group) patients were allowed during the study.

Patient Evaluation

Reduction in PASI score was the primary outcome measure. The PASI score, as proposed originally by Fredriksson and Pettersson [12], was determined initially at base line, at 2 weeks during the next month, and at 4 weekly intervals thereafter. The improvement was graded according to reduction in PASI scores as

shown in Table 1. PASI scores were compared with respective pretreatment baseline values at end of the study and their percentage reduction was analyzed statistically using paired *t*-test and independent *t*-test. A '*p*' value less than 0.05 calculated at 5% level (95% confidence limits) was considered statistically significant.

The study design is registered with Central Trial Registry (CTRI: REF/2013/05/005087).

Ethics

This study was performed on human subjects; thus, all patients were aware of the presence of the study and they were fully informed about the drug and its side-effects.

RESULTS

Baseline demographic and disease severity scores of 43 (M: F 30:13) patients are shown in Table 2. The majority, 27(62.8%) patients were aged between 31 and 60 years. The PASI score ranged between 6 and 22.8 (mean 12.15±5.15) and overall duration of psoriasis was 6 months to 24 (mean 8.59±7.33) years. Two patients from Group-B opted for methotrexate treatment due to disease exacerbation or un-satisfaction with treatment response. Other 16 patients dropped out at various stages of the study (Fig. 1). Baseline characteristics of 25 (M: F 17:8) patients, 14 patients in Group-A and 11 patients in Group-B, who completed the study are shown in Table 3.

Both methotrexate and isotretinoin were effective in the treatment of psoriasis (Figs 2 and 3). Overall 92% of all patients, 14 patients in Group-A and 9 patients in Group-B, had reached the threshold for a minimal response which means a 25% reduction from baseline in PASI score after 12 weeks of treatment. The mean percentage reduction in the PASI scores of patients from the respective pretreatment levels in both groups was also statistically significant ($p < 0.05$) at each follow up at 2, 4, 8, and 12 weeks and improved progressively as the treatment progressed (Fig. 4, Table 4). Methotrexate was more effective than isotretinoin at all follow up visits as the difference in percentage reduction in PASI score of both groups was statistically significant ($p < 0.05$) at the end of 2 weeks, 4 weeks, 8 weeks and 12 weeks. At the end of 2 weeks 8 (57.14%) patients in Group-A showed mild improvement while 5 (35.71%) patients were non-responders. However, at the end of four weeks 9 (64.29%) patients showed moderate improvement and 4 (28.57%) patients had mild improvement. Seven (50%) patients continued

Table 1: Grades of improvement by reduction in PASI Score

Grades of improvement	Reduction in PASI score (%)
Almost complete remission	≥90
Marked improvement	≥75 – 90
Moderate improvement	≥50 – 75
Mild improvement	≥25 – 50
Non responder	<25

Table 2: Pretreatment baseline characteristics of all enrolled patients

Baseline characteristics	Total (*n*=43)	Group-A (*n*=22)	Group-B (*n*=21)
Sex (M:F)	30:13	17:5	13:8
Age (years)			
Range	19-72	23-70	19-72
Mean	45.51±15.16	46.45±12.82	44.52±17.54
Duration of disease (years)			
Range	0.5-24	0.5-24	0.6-23
Mean	8.59±7.33	9.1±7.35	8.7±7.43
Pre treatment PASI score			
Range	6-22.8	6.6-22.8	6-20.5
Mean	12.15±5.15	12.59±5.4	11.69±4.97

Table 3: Pretreatment baseline characteristics of the patients who completed the study

Baseline characteristics	Total (*n*=25)	Group-A (*n*=14)	Group-B (*n*=11)
Sex (M:F)	17:8	12:2	5:6
Age (years)			
Range	23-65	23-65	23-64
Mean	46.80±12.82	48.93±11.61	44.09±14.31
Duration of disease (years)			
Range	0.5-23	0.5-20	0.5-23
Mean	8.46±6.55	8.39±6.4	8.55±7.06
Pre treatment PASI score			
Range	6-22.8	7-22.8	6-20.5
Mean	11.78±5.04	12.21±5.24	11.25±4.96

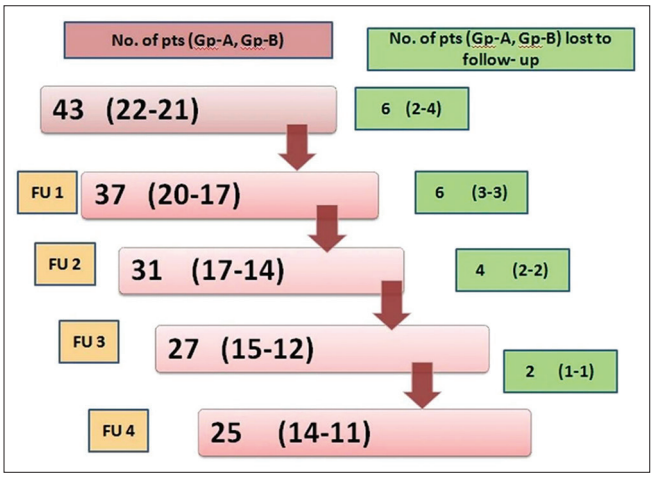


Figure 1: Number of patients at each visit and dropouts (Notes: FU - follow up; Gp-A = Group-A; Gp-B =Group-B).

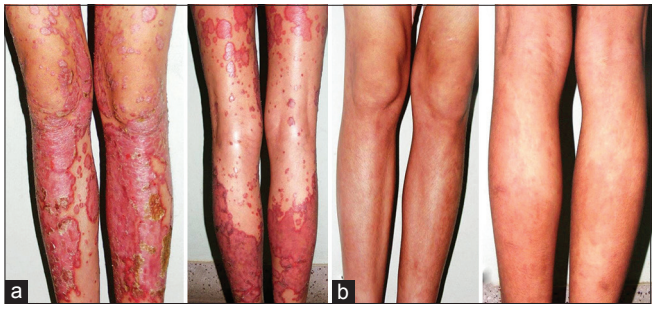


Figure 2: A Group-A patient (methotrexate treatment, 15mg/week). (a) Before treatment, (b) 12 weeks after treatment.

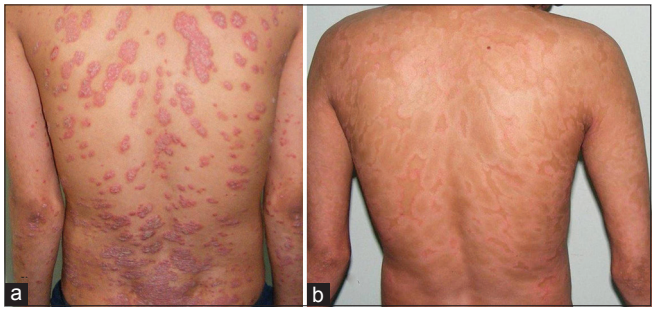


Figure 3: A Group-B patient (isotretinoin treatment, 20mg/d). (a) Before treatment, (b) 12 weeks after treatment.

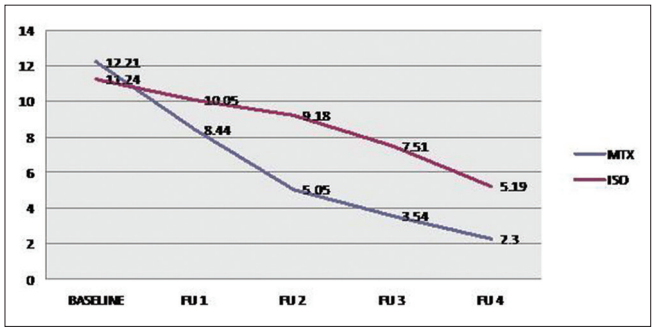


Figure 4: Mean PASI scores of patients who completed the study. (Notes: FU1 = follow up at 2 weeks, FU2 = follow up at 4 weeks, FU3 = follow up at 8 weeks, FU4 = follow up at 12 weeks. MTX = methotrexate (Group-A) and ISO = isotretinoin (Group-B).

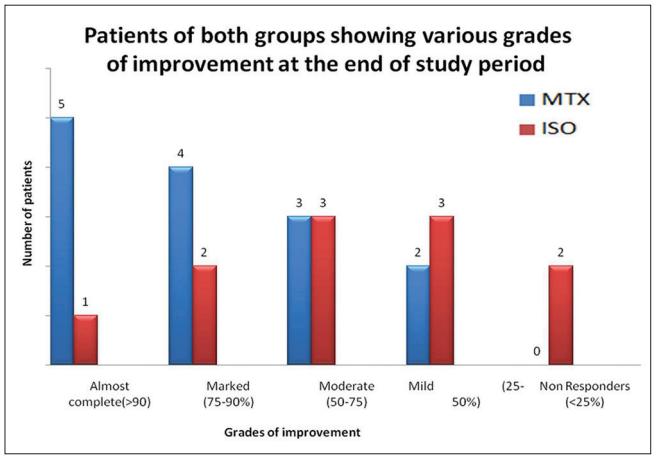


Figure 5: Number of patients having different grades of improvement in both groups [Notes: MTX = methotrexate (Group-A) and ISO = isotretinoin (Group-B)].

Table 4: Characteristics of the patients who completed the study

Characteristics	Group-A (*n*=14)	Group-B (*n*=11)	*p*-value
Sex (M:F)	12:2	5:6	
Age (years)			
Range	23-65	23-64	
Mean	48.93±11.61	44.09±14.31	
Duration of disease (years)			
Range	0.5-20	0.5-23	
Mean	8.39±6.4	8.55±7.06	
Pre treatment PASI score			
Range	7-22.8	6-20.5	
Mean	12.21±5.24	11.25±4.96	
Mean reduction in PASI score at			
2 weeks	8.44±3.65	10.05±4.81	
4 weeks	5.05±2.5	9.18±4.29	
8 weeks	3.54±2.36	7.50±3.62	
12 weeks	2.3±2.09	5.19±2.87	
Mean percentage reduction in PASI score at			
2 weeks	30.64±14.43	11.37±11.67	0.002
4 weeks	57.90±19.30	16.25±28.16	<0.01
8 weeks	70.04±20.62	29.23±33.56	<0.01
12 weeks	79.78±20.68	51.92±23.83	<0.01

Notes: Psoriasis area and severity index (PASI) score of 6 to 12 and involvement of minimum 10% body surface was taken as moderately severe psoriasis. At end of 12 weeks, 5 patients in Group-A achieved >75% reduction in the PASI score, while only 3 patients in Group-B showed similar results. The results were statistically significant ($p<0.05$)

to improve until end of 8 weeks. At the end of the study 4 (28.57%) patients had marked improvement and 5 (35.71%) patients showed complete remission. Ten (90.9%) patients in Group-B were non-responders initially at 2 weeks but 4 (36.36%) patients among these showed mild improvement at 4 weeks. At the end of 8 weeks mild improvement or no response was observed in 4 (36.36%) patients each. However, at the end of study marked to complete remission, moderate improvement or mild improvement occurred in 3(27.27%) patients each (Fig. 5).

Side Effects

Adverse effects from drugs were also noted in both the groups. Eight (57.14%) patients in Group-A experienced methotrexate induced nausea, vomiting, retching and/or anorexia from first month to end of the study period and were managed with antiemetics. Mild abnormality of liver enzymes and thrombocytopenia in one patient each at the end of 8 weeks warranted its discontinuation. However, the patient developing thrombocytopenia had already achieved PASI 75 before discontinuation of methotrexate. Ten (90.9%) patients in Group-B experienced mucocutaneous adverse effects like chelitis, dryness of skin and mouth. One patient complained epigastric discomfort and 4 patients showed altered lipid profile. None of the

patient required discontinuation of treatment due to side effects.

DISCUSSION

Psoriasis affects both the genders with some predilection for male but the result vary according to sampling methodology and most studies exclude females [13-15]. Although women may have an early onset, the age of onset varies across reported studies and bimodal distribution has been reported between 16-22 and 57-60 years of age. However, the disease has no phenotypic difference in either sex. Our 43 patients comprised 30 males and 13 females aged 19-72 (mean 45.51 ± 15.16) years and their demographic profile in both the groups were comparable between them and with the reported studies [15-20]. The fewer number of female patients in this study is perhaps for the simple reason of their exclusion during child-bearing age from such studies, small sample size or perhaps that fewer women seek medical treatment at early stage of their disease.

Methotrexate has become gold standard in treatment of psoriasis despite concerns for hepatic toxicity/fibrosis. A plethora of literature exists on its superior efficacy in psoriasis [13-26]. Haustein et al [24] in a 26-year retrospective study had 157 patients with extensive plaque psoriasis or erythrodermic, pustular and arthropathic forms treated with low-dose methotrexate (15-20 mg/week) for long-term periods. The effect of methotrexate was good in 76% patients, moderate in 18% patients and poor in 6% patients and only 20% of cases discontinued the therapy due to side effects. Griffiths et al [27] suggested that methotrexate reduces the severity of psoriasis by at least 50% in at least 75% of patients. Kumar et al [25] reviewed 244 psoriatic patients treated with weekly oral methotrexate from 1981 to 2000 and observed more than 75% improvement in 88% patients in 8.5±5.1 weeks. Weinstein et al [13] and Sandhu et al [17] observed more than 75% improvement in PASI score at the end of 2-3 months and 12 weeks' treatment with methotrexate. Similarly, Heydendael et al [16] observed complete remission (>90% reduction in baseline PASI score) in their 40% patients and partial remission (>75 % reduction in their baseline PASI score) in 60% patients. The mean pretreatment PASI score in our Group-A patients was 12.21±5.24 and at the end of the study 35.71% patients achieved almost complete remission, 64.29% patients achieved PASI 75 and 85.71% patients achieved PASI 50. The mean percentage reduction in PASI score of 79.78±20.68 after 12 weeks of treatment is comparable to other studies [15,17,19]. Although isotretinoin is considered more effective in pustular psoriasis than in plaque psoriasis [9,10,27,28], there are no studies comparing its efficacy with methotrexate in psoriasis. Anecdotal reports on its use in chronic plaque psoriasis suggest its efficacy as well. Moy et al [10] compared isotretinoin with etretinate in chronic plaque psoriasis and observed moderate to complete response in 4 of 10 patients treated with isotretinoin (1.5 mg/kg/day x 8 weeks). However, isotretinoin has shown equal efficacy to other retinoids when combined with PUVA [29]. The mean pretreatment PASI score in our Group-B patients was 11.25±4.96. The mean percentage reduction in PASI score at the end of 12 weeks was 51.92±23.83 and 3 (27.27%) patients achieved marked to complete remission. Isotretinoin appeared less effective in the first 8 weeks and 4 (36.36%) patients had either mild improvement or were non responder in this study. Overall, the mean percentage reduction of PASI score in our study was higher in methotrexate group than in isotretinoin group at each visit that was statistically significant.

Side Effects

Gastro intestinal (5 patients), mucocutaneous (2 patients), altered liver enzymes (1patient) and thrombocytopenia (1patient) are well known side effects of methotrexate and were observed in 8 (57.14%) patients in Group-A. Flytstom et al [18] observed gastrointestinal side effects as most common adverse effect. van Dooren-Grebbe et al [14] reported methotrexate related side effects (abnormal liver function tests in 44%, nausea in 43%) in their 73% patients. Similarly, Heydendael et al [16] reported nausea in 19(44.19%) patients among 29 (67.44%) patients having gastrointestinal adverse effects. Akhyani et al [17] also observed nausea (80%) and altered liver function test (33.3%) in majority of their patients. Arani et al [20] noted flushing in 2 patients and influenza like symptoms in 7 patients on methotrexate and had to stop methotrexate in 4/25 patients due to liver function abnormalities and recurrent angina. Only in one of our patients, who had marked improvement, treatment was stopped due to thrombocytopenia that recovered after stopping methotrexate. Except for cheilitis (10 patients), altered lipid profile (4 patients) and dry skin/mouth (1 patient each), the well reported

adverse effects of isotretinoin, no serious adverse drug reactions warranting discontinuation of treatment were observed in Group-B.

The high dropout rate in the present study was perhaps due to long distance travel for follow up as has been reported often by these patients. Moreover, psoriasis being a chronic disease adherence to treatment or satisfaction level of these patients remains low despite repeated counseling for continuation of therapy and regular follow up. Other reasons for high dropout rate in Group-B were perhaps cost, low efficacy and slow onset of action of isotretinoin as compared to methotrexate.

CONCLUSIONS

Isotretinoin has been used to treat pustular psoriasis primarily and appears to be effective in at least some patients with moderate-to-severe plaque psoriasis. It has fewer serious adverse effects than methotrexate. However, methotrexate is more effective and leads to a faster clearance of the disease as compared to isotretinoin. Isotretinoin may perhaps be one of the options for alternative therapy in patients with mild disease, or who are intolerant to methotrexate, have achieved its cumulative dose, or in rotational therapy when other options are unaffordable, unavailable, or not tolerated. However, better-planned trials are needed to draw any definite conclusion, find full potential of this mode of treatment and devise a standard treatment protocol.

Limitations

It was an un-blinded study and many patients of Group-B had previous history of methotrexate intake. The dose of isotretinoin (30mg/d equivalent to 0.75mg/kg/d for patient averagely weighing 40kg) in this study was at lower end of 0.75-1.5 mg/kg/d used by Moy et al [10]. Possible spontaneous remissions without treatment, small number of patients and high dropout rate of patients at various stages of the study are some of the other limitations to make any recommendation.

Statement of Human and Animal Rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of Informed Consent

Informed consent was obtained from all patients for being included in the study.

REFERENCES

1. Griffiths CEM, Barker JNWN. Psoriasis. In: Burns T, Breathnach S, Cox N, Griffiths C, editors. Rook's Textbook of Dermatology. 8th ed. Blackwell Publishing: Oxford; 2010.P.20.1-20.60.
2. Gudjonsson JE, Elder JT. Psoriasis. In: Wolff K, Goldsmith LA, Katz SI, Gilchrest BA, Paller AS, Leffell DJ, editors. Fitzpatrick's Dermatology in General Medicine. 7th ed. Mc Graw Hill: New York; 2008.P.169-94.
3. Joshi R. Immunopathogenesis of psoriasis. Indian J Dermatol Venereol Leprol. 2004;70:10-2.
4. Aaronson DS, Lebwohl M. Review of therapy of psoriasis: the prebiologic armamentarium. Dermatol Clin. 2004;22:379-88.
5. Roenigk HH, Auerbach R, Malibach H, Weinstein G, Lebwohl M. Methotrexate in psoriasis: consensus conference. J Am Acad Dermatol. 1998;38:478-85.
6. Kalb RE, Strober B, Weinstein G, Lebwohl M. Methotrexate and psoriasis: 2009 National Psoriasis Foundation Consensus Conference. J Am Acad Dermatol. 2009;60:824-37.
7. Arechalde A, Saurat J-H. Management of psoriasis: the position of retinoid drugs. Biodrugs. 2000;13:327-33.
8. Marhold I, Duschet P, Schwarz T, Gschnait F. Successful use of isotretinoin in Zumbusch generalized pustular psoriasis following recovered etretinate-induced hepatitis. Hautarz. 1991;42:580-3.
9. Sofen HL, Moy RL, Lowe NJ. Treatment of generalised pustular psoriasis with isotretinoin. Lancet. 1984;1:40.
10. Moy RL, Kingston TP, Lowe NJ. Isotretinoin vs etretinate therapy in generalized pustular and chronic psoriasis. Arch Dermatol. 1985;121:1297-301.
11. Feldman SR. A quantitative definition of severe psoriasis for use in clinical trials. J Dermatolog Treat. 2004;15:27–9.
12. Fredriksson T, Pettersson U. Severe psoriasis-oral therapy with a new retinoid. Dermatologica. 1978;157:238–44.
13. Weinstein GD, Frost P. Methotrexate for psoriasis: a new therapeutic schedule. Arch Dermatol. 1971:103:33-8.
14. van Doore-Greebe RJ, Kuijpers ALA, Mulder J, Boo TD, van de Kerkhof PCM. Methotrexate revisited effects of long-term treatment in psoriasis. Br J Dermatol. 1994;130:204-10.
15. Ranjan N, Sharma NL, Shanker V, Mahajan VK, Tegta GR. Methotrexate versus hydroxycarbamide (hydroxyurea) as a weekly dose to treat moderate-to-severe chronic plaque psoriasis: A comparative study. J Dermatolog Treat. 2007;18:295-300.
16. Heydendael VMR, Spuls PI, Opmeer BC, de Borgie CAJM, Reitsma JB, Goldschmidt WFM, et al. Methotrexate versus cyclosporine in moderate to severe chronic plaque psoriasis. N Eng J Med. 2003;349:658-65.
17. Sandhu K, Kaur I, Kumar B, Saraswat A. Efficacy and safety of cyclosporine versus methotrexate in severe psoriasis: a study from north India. Indian J Dermatol. 2003;30:458-63.
18. Flystorm I, Stenberg B, Svensson A. Methotrexate vs cyclosporin in psoriasis: effectiveness, quality of life and safety. A randomised controlled trial. Br J Dermatol. 2008;158:116-21.
19. Akhyani M, Chams-Davatchi C, Hemami MR, Fateh S. Efficacy and safety of mycophenolate mofetil vs. methotrexate for the treatment of chronic plaque psoriasis. J Eur Acad Dermatol Venereol. 2010;24:1447–51.
20. Arani SF, Neumann H, Hop WCJ and Thio HB. Fumarates vs. methotrexate in moderate to severe chronic plaque psoriasis: a multicentre prospective randomized controlled clinical trial. Br J Dermatol. 2011;164:855-61.

21. Bishnoi P, Kumari R, Thappa DM. Monitoring methotrexate hepatotoxicity in psoriasis. Indian J Dermatol Venereol Leprol. 2011;77: 545-8.
22. Boffa MJ. Methotrexate for psoriasis: current European practice. A postal survey. J Eur Acad Dermatol Venerol. 2005;19:196.
23. van de Kerkhof PCM, Mali JWH. Methotrexate maintenance following Ingram therapy in 'difficult' psoriasis. Br J Dermatol. 1982;106:623-7.
24. Haustein UF, Rytter M. Methotrexate in psoriasis: 26 years experience with low dose long-term treatment. J Eur Acad Dermatol Venerol. 2000;14:382-8.
25. Kumar B, Saraswat A, Kaur I. Short-term methotrexate therapy in psoriasis: a study of 197 patients. Int J Dermatol. 2002;41:444-8.
26. Naldi L, Griffiths CEM. Traditional therapies in the management of moderate to severe psoriasis: an assessment of benefits and risk. Br J Dermatol. 2005;152:597-615.
27. Griffiths CEM, Clark CM, Chalmers RJG, Li Wan Po A, Williams HC. A systematic review of treatments for severe psoriasis. Health Technol Assess. 2000;4:1-133.
28. Fry L. Psoriasis. Br J Dermatol. 1988;119:445-61.
29. Halverstam CP, Lebwohl M. Non standard and off-label therapies for psoriasis. Clinic Dermatol. 2008;26:546-53.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Brief Report

Study of basal cell carcinoma and its histopathological variants

Shivanand Gundalli[1], Rutuja Kolekar[2], Amit Kolekar[3], Vikrant Nandurkar[4], Kaveri Pai[5], Sunita Nandurkar[6]

[1]Department of Pathology, S.Nijalingappa Medical College, Bagalkot, Karnataka, India, [2]Department of Obstetrics and Gynaecology, S.Nijalingappa Medical College, Bagalkot, Karnataka, India, [3]Department of Surgery, ABVRH, Sawangi, Wardha, Maharashtra, India, [4]Department of Surgery, Devraj urs Medical College, Kolar, Karnataka, India, [5]Department of Anaesthesia, ABVRH, Sawangi, Wardha, Maharashtra, India, [6]Department of Obstetrics and Gynaecology, Devraj urs Medical College, Kolar, Karnataka, India

Corresponding author: Dr. Shivanand Gundalli, E-mail: drsmgundalli@gmail.com

ABSTRACT

Introduction: Basal Cell Carcinoma (BCC) typically affects older individuals with Predilection for sun-exposed skin (face, hands) small, well-circumscribed, pearly tan-gray papule devoid of scale lesions enlarge with time and tend to ulcerate (rodent ulcers). Basal cell nevus syndrome: multiple basaloid hamartomas on the cutaneous surface associated with palmar keratotic pits, jaw cysts, and basal cell carcinomas in non–sun-exposed locations. BCC rarely metastasize; when they do, the primary lesion is usually advanced. **Aim:** The aim of our study was to determine the frequency of various types of BCCs encountered in our practice, to delineate the spectrum in our setup and to determine the different histological patterns, anatomical location, site predilection, and age and sex incidence. **Materials and methods:** The present study was carried out in the department of pathology in a tertiary care centre. The study was prospective (2years) as well as retrospective (5 years) and was done during the period of September 2004 to September 2011 i.e., 7 years. **Results :** In the present study 21 cases of BCC were seen and accounted for 26.25% of all the malignant tumors of skin. **Conclusions :** In the present study majority (85%) of the lesions of BCC were located on head and neck region, average age of cases of basal cell carcinoma was 65.6 years, and solid type of BCC was most common type.

Key words: Basal cell carcinomas; Study; Age

INTRODUCTION

The first description of basal cell carcinoma (BCC) was by Jacob in 1827. BCC is the most common malignant skin tumor, but the incidence in Asian races is lower than in the white race. The incidence of BCC in Indian literature ranges from 12% to 30%.BCC is seen almost exclusively on hair bearing skin especially on the face. BCC generally occurs in adults. BCC may develop in children under following circumstances: Nevoid basal cell epithelioma syndrome, in pre-existing organoid nevus and in xeroderma pigmentosa [1].

There are five clinical types of Basal cell carcinoma which include [2]

1. Nodulo-ulcerative
2. Pigmented
3. Morphea-like or fibrosing Basal cell carcinoma
4. Superficial (multifocal)
5. Fibroepithelioma of Pinkus.

Scriverer Y et al in his study on 13,457 patients with BCC have shown that nodular and morphoeiform types predominate on the forehead where as the trunk is the most common site for the superficial type [3].

Histopathology: BCC tend to share the common features of a predominant basal cell type, peripheral palisading of cell nuclei, a specialized stroma, and clefting artifact between the epithelium and the stroma.

How to cite this article: Gundalli S, Kolekar R, Kolekar A, Nandurkar V, Pai V, Nandurkar S. Study of basal cell carcinoma and its histopathological variants. Our Dermatol Online. 2015;6(4):399-403.

Submission: 05.03.2015; **Acceptance:** 01.06.2015

DOI: 10.7241/ourd.20154.107

Prognosis and predictive factors: BCC are locally invasive tumours and metastases occur in less than 1 in 10,000 tumours. Morbidity is increased with deeply invasive tumours which may extend into the deep tissue to bone and follow fusion planes particularly on the face where they follow nerves through bony channels. Morbidity also increases with neglected tumours that may measure more than 10 cm in diameter and have been described as giant basal cell carcinomas. Multiple recurrences with deep residual tumour on the head may be associated with particular morbidity as basal cell carcinomas can ultimately penetrate the cranium. Increased recurrences are associated with infiltrative, morphoeic and micronodular BCC as surgical margins may be underestimated. BCC recurrences are more common in lesions on the nose and nasolabial fold, this may be in part due to the difficulty in achieving adequate margins in these sites. Tumours recurring after radiotherapy are usually aggressive and infiltrative [4].

MATERIALS AND METHODS

The present study was carried out in the department of pathology in a tertiary care centre. This study included tumours of epidermis along with melanocytic tumours and adnexal tumours of skin including secondaries without restricting the study to any particular age limit. Mesenchymal tumours of skin, haematological tumours of skin, neural tumours of skin, nonneoplastic lesions of skin and all tumours arising from mucosal area of mucocutaneous junction such as glans penis and eyelid margin were excluded. The study was prospective (2years) as well as retrospective (5 years) and was done during the period of September 2004 to September 2011 i.e., 7 years. Data for retrospective study was obtained from departmental records, tissue blocks and slides. Data for prospective study was obtained from clinical records, tissue specimens, tissue blocks and slides Clinical details were obtained and maintained according to the proforma. All the biopsies and resected specimens received in the histopathology section were immediately fixed in 10% formalin for 24 hours. Gross features of the specimen were noted. Multiple sections of the specimen were taken. Then they were processed and embedded in paraffin wax. Three-five microns thick sections were prepared and then stained with Haematoxylin & Eosin. Detailed study of the sections was performed under the light microscope and then the final diagnosis was given.

Ethical clearance: Ethical clearance has been obtained from Ethical committee of institution. Statistical methods applied:

Following Statistical methods were applied in the present study.

1. Number and percentage
2. Descriptive statistics.

Prior to the study, every patient gave written consent to the examination and biopsy after having been informed about the procedure and aim of the study.

RESULTS

In the present study 21 cases of BCC were seen and accounted for 26.25% of all the malignant tumours of skin (Figs 1 – 6). Maximum number of cases were observed in 7th decade. Mean age of patients was 65.6 years.

Table 1 shows sex distribution of BCC. This table shows predominance of BCC in males with male to female ratio 1.3:1.

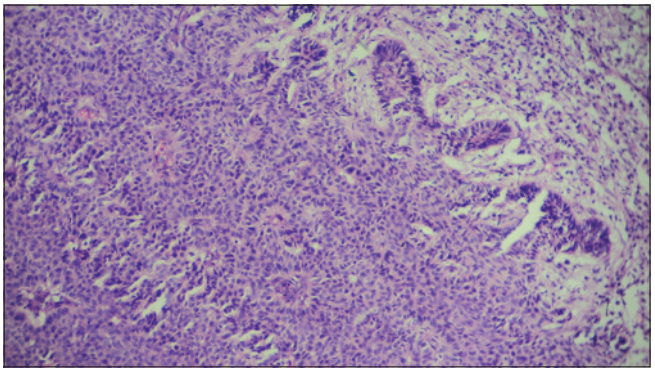


Figure 1: Adenoid bcc. Formations suggesting tubular, gland-like structures. 100x,H&E.

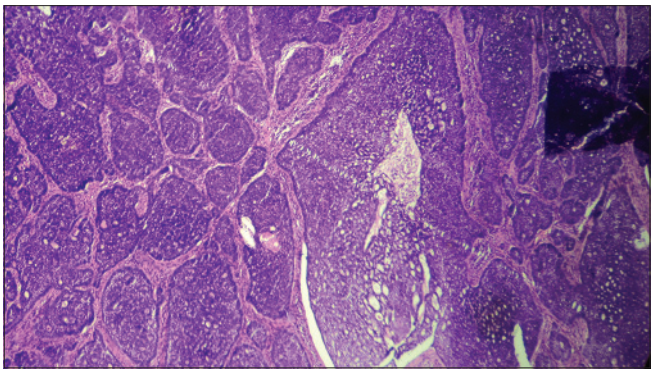


Figure 2: Basal Cell Carcinoma. Solid Circumscribed. Tumor masses of various sizes and shapes embedded in the dermis. The peripheral cell layer of the tumor masses often shows a palisade arrangement. 100x,H&E.

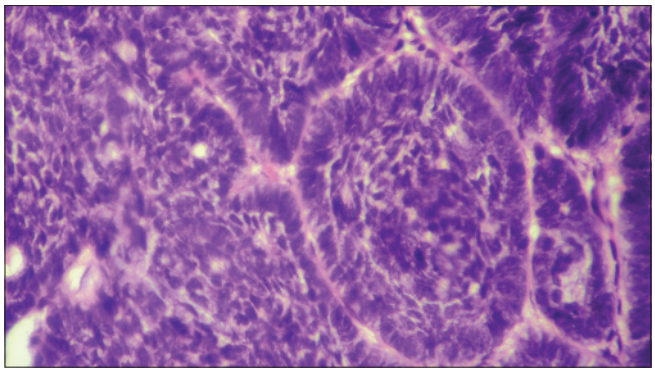


Figure 3: Basal Cell Carcinoma. Solid Circumscribed. The peripheral cell layer of the tumor masses often shows a palisade arrangement and Artefactual retraction spaces between the tumour and stroma are often present. 400x,H&E.

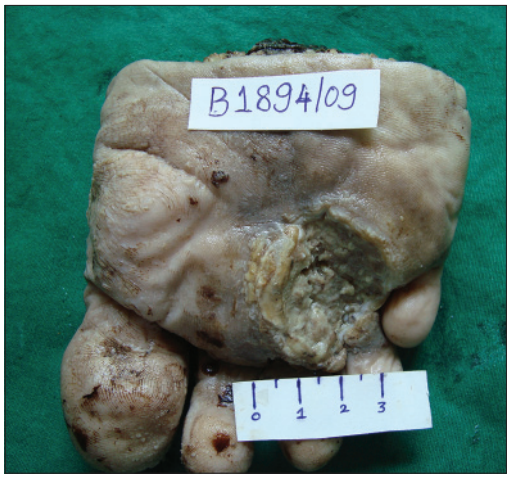


Figure 4: Noduloulcerative (rodent ulcer) Basal cell carcinoma: Photomicrograph showing central ulceration in amputated foot region which is confirmed after histopathogy.

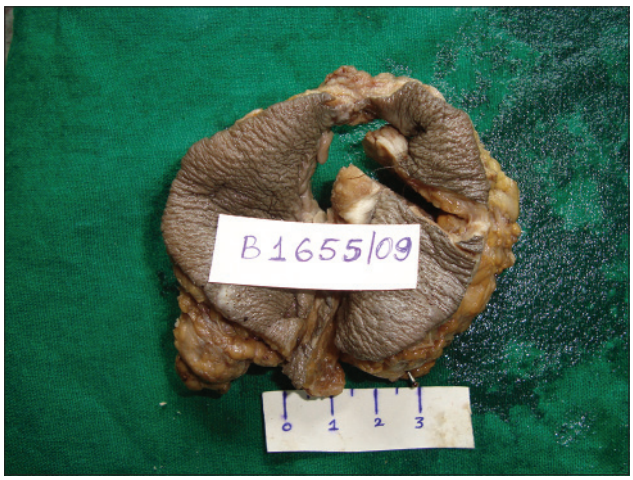


Figure 5: Noduloulcerative (rodent ulcer) Basal cell carcinoma: Photomicrograph showing slowly enlarging ulcer surrounded by a pearly, rolled border in head and neck region.

Table 2 show site wise distribution of BCC. As shown in this table, 85% of cases of BCC were located on head and neck region and majority were seen in the eye lids.

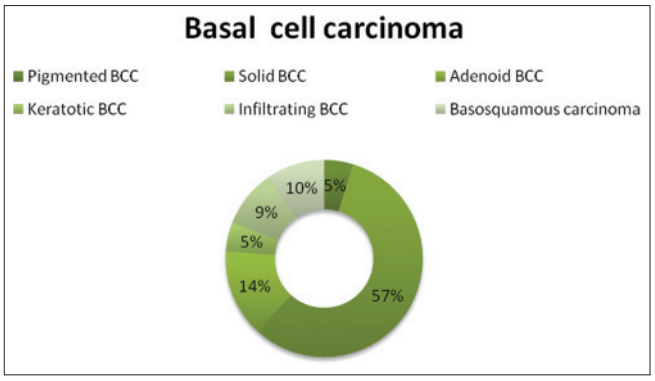


Figure 6: Histological types of basal cell carcinoma.

Table 1: Sex distribution of basal cell carcinoma

Sex	Number of cases	Percentage
Males	12	57.2
Females	9	42.8
Total	21	100

Table 2: Site wise distribution of basal cell carcinoma

Site	Number of cases	Percentage
Head and neck		
Eye lid	6	28.58
Nose	5	23.82
Forehead	2	9.52
Cheek	2	9.52
Ear front	1	4.76
Scalp	1	4.76
Post auricular	1	4.76
Trunk	2	9.52
Limb	1	4.76
Total	21	100

In figure 4 we presented histological types of BCC. As shown in the above chart, in the present study solid variant of BCC was the commonest (57.14%) histological type followed by adenoid type, infiltrating type and basosquamous type. BCC with no differentiation were categorized as solid basal cell carcinoma. Adenoid BCC showed cellular arrangement suggesting tubular structures. The cells were arranged in intertwining strands resulting in a lacelike pattern. Keratotic basal cell carcinoma showed keratinisation in the form of horn cysts. In the infiltrating variant of BCC the basaloid cells were arranged in cords and showed deep infiltration in the dermis. Two cases of basosquamous carcinoma were found in the present study. Both of them showed combined features of basal cell carcinoma and squamous cell carcinoma. Both cases were females of 63 and 65 years age presented as ulcer over the scalp and ulcer on left nostril.

Table 3 show age distribution of malignant tumours of skin compared with BCC. As shown in this table maximum number of cases that is 63 (78.75%) among 80 malignant BCC sincluding secondaries belonged to the age group of 6th to 8th decade.

DISCUSSION

Frequency of occurrence of BCC with respect to malignancies of skin in various other studies show table 4.

In the present study the frequency of occurrence of basal cell carcinoma was 26.25% of all the malignant tumours of skin. The frequency of occurrence of basal cell carcinoma in other studies ranged from 18% to 30%. Our findings are comparable to others studies.

In the present study majority (85%) of the lesions of BCC were located on head and neck region, which is similar to the observations of Solanki et al [5] (94%), Chakravorthy et al [6] (90%) and Budhraja et al [7] (78%).

In the present study male to female ratio in the patients of BCC was 1.33:1. Solanki, et al found a male to female ratio of 1.26:1. Hence our findings are comparable with Solanki, et al [5].

And in present study the average age of cases of BCC was 65.6 years. Scrivener, et al noted average age as 65 years. Hence our findings are comparable to Scrivener, et al [3].

Table 5 shows comparison of histological types in BCC.

In the present study solid type of BCC was most common type which is comparable to the study of Solanki et al [5] as he also noted the solid type as most common type of BCC in his study of 172 cases.

BCCs constitute a small but significant proportion of patients with cancer. The skin is a complex organ. Because of its complexity a wide range of diseases develop from the skin including tumors from surface epidermis, epidermal appendages and dermal tissue.

The diagnosis of BCCs presents unique difficulties, in part, related to the wide variety of tumors and the complicated nomenclature. The study of histogenesis of the basal cell carcinomas is interesting, fascinating and challenging because of wide range of differentiation.

Histopathological study is one of the most valuable means of diagnosis in dermatopathology and the diagnosis of BCCs can be done by correlating clinical features, gross and histological appearances.

The present study emphasizes the various patterns of skin neoplasms in this geographic location in and around city.

Finally the quintescence of the subject of study of basal cell carcinomas is it's vastness, it's enormity and its interesting histomorphology.

CONCLUSIONS

In the present study majority (85%) of the lesions of BCC were located on head and neck region, average

Table 3: Age distribution of malignant tumours of skin compared with basal cell carcinoma

Malignant tumours	0-9	10-19	20-29	30-39	40-49	50-59	60-69	70-79	80-89	90-99	Total
SCC	-	-	2	1	2	7	16	8	1	-	37
Verrucous Carcinoma	-	-	-	1	1	1	1	-	-	-	4
BCC	-	1	-	-	3	4	8	4	1	-	21
Malignant melanoma	-	-	-	1	1	1	4	1	-	1	9
Adnexal carcinoma	-	-	-	-	1	2	3	-	-	-	6
Secondaries	-	-	-	-	-	-	2	1	-	-	3
Total	-	1	2	3	8	15	34	14	2	1	80

Table 4: Comparison of frequency of basal cell carcinoma with respect to malignancies of skin in various other studies

Author	Percentage of basal cell carcinoma
Deo S V et al.[1] (2005) (n=14)	18.10
Chakravorthy R C et al.[2] (1968) (n=19)	18
Paymaster et al.[3] (1972) (n=102)	30
Solanki R L et al.[4] (1989) (n=172)	28
Present study (2011) (n=21)	26.25

Table 5: Comparison of histological types in basal cell carcinoma

Histological pattern of BCC	Solanki et al.[4] (1989) (n=172) (%)	Present study (2011) (n=21) (%)
Solid	60.5	57.14
Adenoid	15.7	14.30
Keratotic	9.3	9.52
Pigmented	6.4	9.52
Basosquamous	3.5	4.76
Infiltrating	-	4.76

age of cases of basal cell carcinoma was 65.6 years, and solid type of BCC was most common type.

Statement of Human and Animal Rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of Informed Consent

Informed consent was obtained from all patients for being included in the study.

REFERENCES

1. Kirkham N. Tumours and cysts of epidermis. In Lever's histopathology of skin. 9th ed. Philadelphia: Lippincott Raven; 2005. p.805-866.
2. Mckee PH and Brenn T. Tumours of surface epithelium. In pathology of skin. Elsevier Mosby, 3rd ed; p1153-1237.
3. Scrivener Y, Grosshans and Cribier B. Variations of BCC according to gender, age, location and histological type. Br J Dermatol. 2002;147:41-7.
4. LeBoit PE, Burg G, Weedon D and Sarasin A. Pathology and genetics of skin tumours. In World health organisation classification of tumours. IARC press. Lyon, 2006.p.1-300.
5. Solanki RL, Anand VK, Gaur SK, Arora HL and Gupta R. Neoplasms of hair follicle. Indian J Dermatol Venereol Leprol. 1989;55:33-7.
6. Charkravorthy RC, Choudhuri DR. Malignant neoplasms of the skin in Eastern India. Indian J Cancer. 1968;5:133-44.
7. Budharaja SN, Pillai VCV, Periyanagam WJ, Kaushik SP and Bedi BMS. Malignant neoplasms of skin in Pondicherry- a study of 102 cases. Indian J Cancer. 1972;284-95.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Brief Report

The role of Bleomycin in management of hypertrophic scars and keloids - A clinical trial

Ravi Reddy[1], Sreekar Harinatha[2], Nithya Raghunath[3]

[1]Department of General Surgery, Vijaynagar institue of Medical College, Bellary, Karnataka, India, [2]Department of General surgery, MVJ Medical College, Bangalore, Karnataka, India, [3]Department of Dermatology, MVJ Medical College, Hoskote, Bangalore, India.

Corresponding author: Dr. Sreekar Hainatha, E-mail: drsreekarh@yahoo.com.

ABSTRACT

Introduction: Keloids and hypertrophic scars remain a nagging problem even with the emergence of multiple modalities in their treatment. The wide range of modalities used for their treatment also point out that no single treatment is definitively superior. **Aims:** The aim of this study was to define the role of Bleomycin and to confirm its effectiveness in the management o keloids and hypertrophic scars. **Settings and Design:** This was a prospective clinical trial involving 20 patients with hypertrophic scars and keloids. Methods and Material: The patients were treated with four monthly injections of Bleomycin and were followed for 6 months after the termination of treatment course. Assessment of the size of keloids and hypertrophic scars was done at the beginning, at the time of stopping the therapy and during the follow-up. **Statistical analysis used:** The response to treatment was divided into the following categories: <25 percent flattening = poor response, 26–50 percent flattening = fair response, 51–75 percent flattening = good response and >75 percent flattening = excellent response. **Results:** Of the twenty patients, 13(65%) showed excellent response, 3(15%) showed good response, 2(10%) showed fair response and 2(10%) showed poor response. There was complete resolution of symptoms in 11 patients (55%) and improvement in the other 9(45%). **Conclusions:** Bleomycin is one of the most effective and safest modality for the treatment of hypertrophic scars and keloids.

Key words: Bleomycin, hypertrophic scars, keloids

INTRODUCTION

Keloids and hypertrophic scars form due to uncontrolled dermoproliferation and are unique to humans. They occur following any kind of injury and lead to considerable cosmetic blemish. The mechanism underlying keloid formation has not been fully defined though studies reveal that the activated fibroblasts and excessive collagen production are responsible for their occurrence. The word is derived from the Greek word *chele*, or crab's claw, to describe the growth of scar into neighbouring skin. Although the hypertrophic scars and keloids share some similarities, there are some differences that suggest that both are distinct from each other. Clinically, keloids extend beyond the original wound, whereas hypertrophic scars tend to remain within the borders of the original wound [1].

Presently, a wide range of treatments have been implemented in treating hypertrophic scars and keloids, including surgical excision, silicon sheets/gels, topical and intralesional corticosteroids, interferon, cryosurgery, radiation, pressure therapy, laser therapy retinoic acid, and other anti-neoplastic drugs. The focus of anti-neoplatic drugs like Bleomycin lies on fibroblasts which are the culprit cells in keloids.

Bleomycin is a polypeptide antibiotic with antitumor, antibacterial, and antiviral activity [2]. Studies have shown that intradermal injections result in

How to cite this article: Reddy R, Harinatha S, Raghunath N. The role of Bleomycin in management of hypertrophic scars and keloids - A clinical trial. Our Dermatol Online. 2015;6(4):404-406.

Submission: 07.02.2015; **Acceptance:** 23.07.2015

DOI: 10.7241/ourd.20154.108

significant improvement in keloids and hypertrophic scars [3,4]. Though the exact mechanism by which Bleomycin acts has not been fully elucidated, studies have shown that it inhibits collagen synthesis in dermal fibroblasts through decreased stimulation by TGF-β1 [4]. Bleomycin also reduces lysyl-oxidase levels in cultures of human dermal fibroblasts in vitro [2]. The effect of Bleomycin in hypertrophic scars and keloids may be from a reduction of collagen synthesis, increased destruction from inhibition of lysyl-oxidase or TGF-β1, or both. Rarely have side effects of intralesional administration of bleomycin, been reported, including hyperpigmentation and dermal atrophy. Systemic side effects of bleomycin, hepatotoxicity, and pulmonary fibrosis, are not of concern with intralesional administration [5]. This study was initiated to confirm the effectiveness of Bleomycin in the treatment of hypertrophic scars and keloids in Indian population.

MATERIALS AND METHODS

The study enrolled 20 patients with keloids and hypertrophic scars after obtaining clearance from institutional review board. Written informed consent was taken from all the patients before the study. We classified hypertrophic scar as a red or dark pink, elevated scar confined to the border of the original surgical incision. A keloid is instead classified as a scar red to brown in colour, very elevated, larger than the wound margins very hard and sometimes painful or pruritic with no spontaneous regression. In each case the maximum dose of Bleomycin used was 2 mL/cm^2 of skin treated at a concentration 1.5 IU/ml and a maximum of 6 mL of undiluted bleomycin was given per session. The injections were given in multiple punctures with an insulin syringe. Up to a maximum of 4 doses were administered at intervals of 1 month. The size of keloids was measured by three different observers using calipers and the mean was deduced for accurate size assessment. The response to treatment was divided into the following categories: <25 percent flattening = poor response, 26–50 percent flattening = fair response, 51–75 percent flattening = good response and >75 percent flattening = excellent response. The incidence of side effects if any was noted. Assessment of the hypertrophic scars and keloids was done at the beginning, at the time of stopping the therapy and during the follow-up. The lesions were measured and followed on monthly basis for 6 months.

Ethics

This study was performed on human subjects; thus, all patients were aware of the presence of the study and they were fully informed about the drug and its side-effects.

RESULTS

Twenty (12 male, 8 female) patients were included in the study. The duration of keloids ranged from 3 to 14 (mean 6.6) years. The mean age of the patients was 34 years. A total of 20 keloid and hypertrophic scars were treated. The involved areas were as follows: shoulder, 3; neck, 2; ear, 4; chest, 8; upper limbs, 2 and face, 1. All the patients had itching, 13 had pain (65%).

Of the twenty patients, 13 (65%) showed excellent response, 3 (15%) showed good response, 2 (10%) showed fair response and 2 (10%) showed poor response (Fig. 1). There was complete resolution of symptoms in 11 patients (55%) and improvement in the other 9 (45%). Almost all patients experienced moderate to severe pain at the injection site, which lasted about 5–15 min which were treated with a single oral analgesic. The drug was given undiluted and in multiple punctures. There was no evidence of hypopigmentation, ulceration or atrophy following injections. All the patients followed up for 6 months after termination of therapy. There were no signs of recurrence or reappearance of the symptoms.

DISCUSSION

Aggressive fibroblasts are the reason for excessive scar formation. Antimitotic drugs selectively inhibit fibroblast proliferation and promote apoptosis, thereby inhibiting scar formation. Some drug treatments, such as steroids,

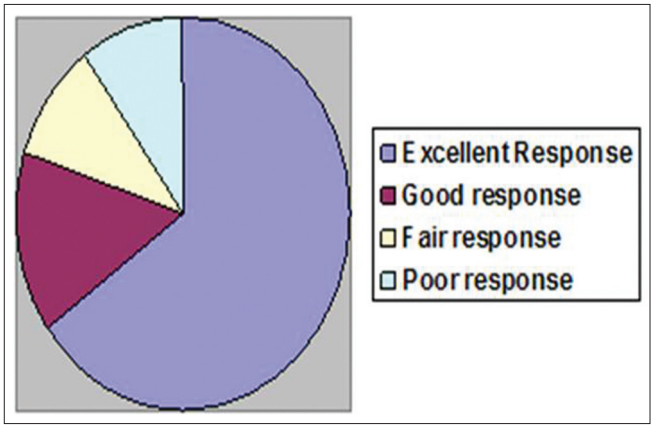


Figure 1. Response (reduction in size) following treatment with bleomycin.

5–FU and Bleomycin have shown positive effects and have potential applications. However, complete resolution for scar is still a difficult problem. Troublesome adverse effects resulting from antimitotic drug injections also limit their application. Presently, numerous methods have been described for the treatment of keloids and hypertrophic scars, but to date, the optimal treatment method has not been established [6].

Bleomycin is a glycopeptide antibiotics, isolated from a strain of Streptomyces verticillus. The main mode of action is inhibition on DNA synthesis and, to lesser extent, inhibition on RNA and protein synthesis. Bleomycin is used as an antitumor agent for treating various malignancies. In addition, bleomycin is used as a treatment of hypertrophic scars, and keloids. Trials have shown that administration of bleomycin by intradermal injections or the multipuncture method is effective against keloid and hypertrophic scars.

The low incidence of side effects makes Bleomycin one of the safest modalities for keloid management. In similar studies, most common complication noted was minor ulceration which healed within 10 days and hyperpigmentation that resolved after 1 year of follow up [7]. None of the patients demonstrated any pulmonary, hepatic, or other major systemic side-effects of bleomycin, which may be in part due to the low dose of Bleomycin used.

Bleomycin usage has a better or comparable result as with other treatment modalities such as Triamcinolone and silicon gel sheets, and has minimal complications and less recurrence rate, which shows its usefulness as the first-line treatment modality for management of keloids and hypertrophic scars.

Statement of human and animal rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of informed consent

Informed consent was obtained from all patients for being included in the study.

REFERENCES

1. Köse O, Waseem A. Keloids and hypertrophic scars: are they two different sides of the same coin? Dermatol Surg. 2008;34:336–46.
2. Yu Z, Schmaltz RM, Bozeman TC, Paul R, Rishel MJ, Tsosie KS, et al. Selective Tumor Cell Targeting by the Disaccharide Moiety of Bleomycin. J Am Chem Soc. 2013;135:2883-6.
3. Naeini FF, Najafian J, Ahmadpour K. Bleomycin tattooing as a promising therapeutic modality in large keloids and hypertrophic scars. Dermatol Surg. 2006;32:1023–9.
4. España A, Solano T, Quintanilla E. Bleomycin in the treatment of keloids and hypertrophic scars by multiple needle punctures. Dermatol Surg. 2001;27:23–7.
5. Shippee BM, Bates JS, Richards KL. The role of screening and monitoring for bleomycin pulmonary toxicity. J Oncol Pharm Pract. 2015 Mar 2.
6. Trisliana Perdanasari A, Lazzeri D, Su W, Xi W, Zheng Z, Ke L, et al. Recent developments in the use of intralesional injections keloid treatment. Arch Plast Surg. 2014;41:620-9.
7. Saray Y, Güleç AT. Treatment of keloids and hypertrophic scars with dermojet injections of bleomycin: a preliminary study. Int J Dermatol. 2005;44:777–84.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Brief Report

Topical corticosteroid abuse on face: A clinical, prospective study

Balwinder Kaur Brar, Kamra Nidhi, Sukhmani Kaur Brar

Department of Dermatology, Guru Gobind Singh Medical College and Hospital, Saadiq Road, Faridkot (Punjab), India

Corresponding author: Dr Nidhi Kamrae, E-mail: nidhikamra06@gmail.com

ABSTRACT

Introduction: Topical corticosteroids are widely misused and uncontrolled use of topical corticosteroids leads to undesirable adverse effects on facial skin. **Aim:** The aim of this study was to assess cutaneous manifestations of topical corcticosteroid abuse on the face and to analyse various factors contributing to the same. **Materials and methods:** A total of 100 patients with facial dermatoses using topical corticosteroids for a minimum period of 1 month attending the outpatient department from a period of January 2013 to September 2014 were enrolled for the study. Details about the usage of topical corticosteroids and their adverse effects were recorded. **Results:** Majority of the patients were females (68%). The most common indication for use was acne (68%), followed by melasma (22%). Most of the patients used them for duration of less than 6 months and most commonly misused steroids were of potent type. The commonest side effect was acneiform eruption (56%) followed by steroid dependent facies (26%). Other adverse effects like hypertrichosis, telangiectasia and premature aged appearance was seen with longer duration of steroid use. **Conclusion:** Misuse of topical corticosteroids is rampant and urgent, quick steps are required to increase the awareness and for better management of its adverse consequences.

Keywords: steroid abuse, acneiform eruptions, steroid dependent facies.

INTRODUCTION

Topical corticosteroids (TCS) are perhaps the most widely used agents amongst the therapeutic armamentarium and have been rightly acknowledged as a wonder drug in dermatological therapy [1]. They provide immediate subjective and objective relief in symptoms in almost all inflammatory dermatoses thus justifying its rampant use. This usefulness of the drug has become a double edged weapon and made alarming proportion of individuals vulnerable to its abuse leading to serious local adverse effects especially on face [2]. Vast sections of society have become victims of this magic drug owing to the craze of beautification leading to a virtual epidemic of acneform eruptions, steroid rosacea/steroid dependent facies, telangiectasia, hypertrichosis and premature aged appearance of face. Aim of our study is to make awareness about misuse of TCS on face and to give brief account of treatment modalities for the same.

METHODS

A total of 100 cases attending the dermatology OPD of GGS Medical College and Hospital from January 2013 to September 2014 were enrolled for the study after taking informed consent. Ethical clearance from the ethical and review committee was taken. Inclusion criteria for patients were the history of use of any TCS on face for duration of more than one month for cosmetic reasons including melasma, acne and non-specific reasons. Patients with history of rosacea, atopic dermatitis, seborrheic dermatitis and contact dermatitis prior to initiation of use of TCS were excluded from the study.

A proper history was taken from patients regarding the duration of use of TCS; type and potency of TCS used; indication for use; source of prescription and symptoms like pruritus, burning, dryness, photosensitivity and rebound phenomenon.

How to cite this article: Kaur Brar B, Nidhi K, Kaur Brar S. Topical corticosteroid abuse on face: A clinical, prospective study. Our Dermatol Online. 2015;6(4):407-410.
Submission: 07.02.2015; **Acceptance:** 26.08.2015
DOI:10.7241/ourd.20154.109

Thorough local cutaneous examination was done including Fitzpatrick skin type (I-VI), site, erythema, scaling, xerosis, telangiectasia, dyspigmentation, atrophy, wrinkling, papules, pustules, nodules and hypertrichosis. The general physical and systemic examination was carried out. Routine laboratory investigations and specific investigations were carried out where indicated and medical photographic documentation was done in all patients.

Patients were instructed to avoid provocative factors (hot beverages, alcohol, spicy food, cosmetic), sun exposure and all the OTC preparations. Treatment options tried according the cutaneous adverse effect and severity were cold saline compresses, bland emollients, physical sunscreens, soap free cleansers, topical calcineurin inhibitors, oral antibiotics like tetracyclines, minocycline and doxycline, oral macrolides, non-cardioselective β-blockers like propranolol and oral vitamin C preparations.

RESULTS

Amongst the 100 patients studied, 68 were females and 32 were males (M: F =1:2.125). Majority of the patients belonged to the age group of 26-35 years (Table 1). Duration of application was less than 6 months in majority, maximum duration being 6 years for melasma in a female. Steroids of different potencies as used by the patients are shown in Table 2. TCS for facial use were prescribed by peers (friends, relatives, beauticians) in 51, chemists in 22, physicians/general practitioners in 23 and by dermatologists in 4. Various indications for which TCS were used are elaborated in Table 3. The adverse effect profile of TCS is shown in Table 4 and Figs. 1-4. We did not observe allergic contact dermatitis to TCS in any of our patients. Amongst the adverse effects hypertrichosis, telangiectasia and premature aged appearance were seen with longer duration of steroid use (>6 months) and with potent steroid use. Majority of the patients with steroid dependent facies gave typical history of flaring of erythema on discontinuation of TCS.

Patients with severe symptoms and steroid dependent facies who were given oral azithromycin 500 mg in the form of weekly pulse therapy (3 tablets per week for 8 weeks) or oral doxycycline/minocycline 100mg once daily for 8 weeks along with β blocker (propranolol 10mg twice a day) and oral vitamin C preparations showed good improvement after 8 weeks. Those with mild symptoms and acneiform eruptions were given topical tacrolimus (0.03%) for 8 weeks and showed good response in 6 weeks. Avoidance of provocative factors, soap free cleansers, physical blockers and emollients were adjuncts to the primary treatment modalities and helped in ameliorating symptoms.

DISCUSSION

The development of TCS in 1950s opened new doors for treatment of intractable dermatoses. In 1970s with introduction of higher potency TCS, new dermatoses related to over application of TCS begin to emerge. The first case of rosacea like dermatitis as a result of abuse of TCS was reported in 1957 [3].

Face is the most common site for TCS abuse owing to rapidity of action and cosmetically appreciable effects. Various terms coined for lesions of steroid abuse are steroid rosacea [4], steroid dermatitis resembling rosacea [5], red face syndrome [2] and steroid dependent facies (SDF). It was observed in 26 patients in our study. The primary lesion in SDF is pinpoint, red or flesh colored papules or pustules. Eventually, patients develop persistent and diffuse erythematous and edematous skin with numerous telangiectatic vessels as well as deep follicular papules, pustules, and firm nodules [6,7]. The

Table 1: Age distribution of patients using TCS on face

15-25	23
26-35	36
36-45	22
46-55	13
>55	6

Table 2: Potency of TCS used

Very potent	18
Potent	40
Moderately potent	34
Mildly potent	8

Table 3: Indications for TCS use

Acne	68
Melasma/fairness product	22
Miscellaneous causes	10

Table 4: Adverse effect profile of TCS use on face

Acneiform eruptions	56
Steroid dependent facies	26
Dyspigmentation	9
Hypertrichosis	4
Telangiectasia	3
Premature aged appearance(atrophy, wrinkling)	2

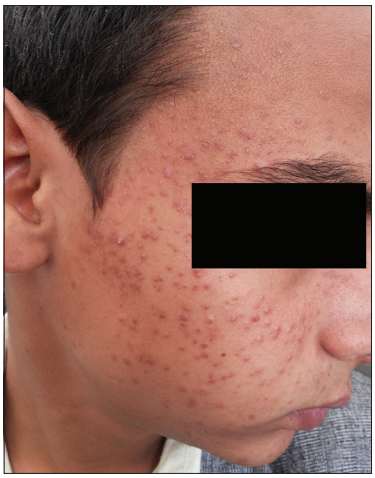


Figure 1: Acneform eruption

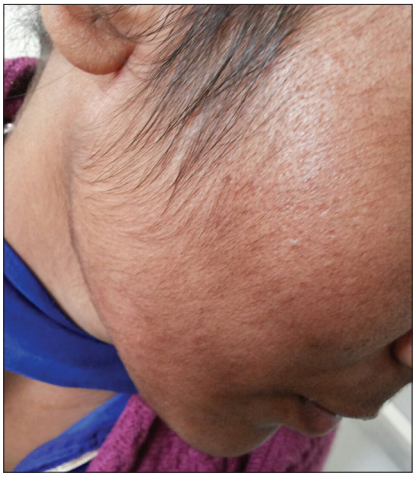


Figure 3: Hypertrichosis

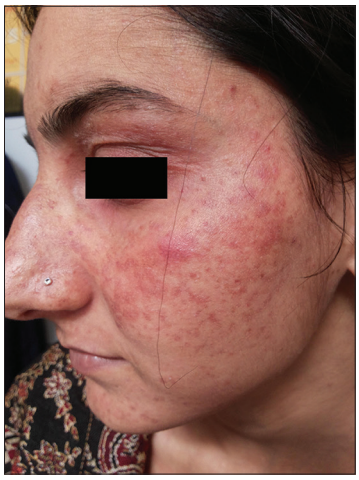


Figure 2: Steroid dependent facies/steroid rosacea

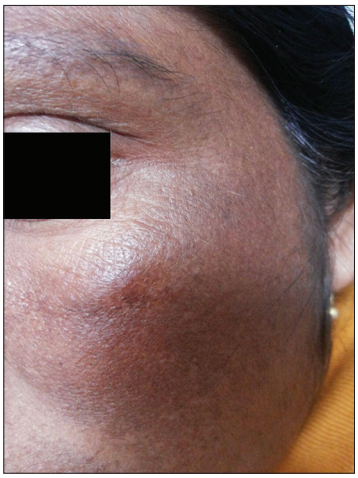


Figure 4: Premature wrinkling

pathogenesis of SDF and its rebound phenomenon is multifactorial. Steroids inhibit the release of endothelium derived relaxing factor. When steroids are withdrawn, the vasoconstrictive effect ceases and blood vessels enlarge leading to erythema, burning and pruritus [8]. The immunosuppressive effect of TCS may facilitate the overgrowth of various bacteria, yeast, demodex mites or other microorganisms, resulting in inflammatory reaction that leads to papules and pustules [9].

Acneiform eruptions are also the common side effect observed with use of TCS and was seen in 56 patients in our study. Bhat Y et al observed SDF more than acneiform eruptions in his study [10]. It was interesting to observe that majority of our patients used TCS for various forms of inflammatory acne. Initially majority of the patients experienced relief but on prolonged use developed monomorphic, pigmented papular or nodular lesions.

Other adverse effects observed in our study were telangiectasia, hypo/hyper-pigmentation, hypertrichosis and premature aged appearance as a result of atrophy and wrinkling. They were observed less commonly as in other studies. [11] None of our patients reported allergic dermatitis to TCS or peri-oral dermatitis. Most of the allergic reactions reported due to use of TCS have been attributed to the base [12].

Our study showed a good response to the combination of oral antibiotics (azithromycin and tetracycline group) and propranolol in severe cases and topical tacrolimus 0.03% in mild cases. Oral antibiotics were used in subantimicrobial doses owing to their anti-inflammatory properties [13]. Beta-blockers antagonize the flushing reaction and hence were used in severe

cases. Tacrolimus exerts its immunosuppressive and anti-inflammatory effects by blocking T cell activation. However in contrast to TCS it does not induce vasoconstriction and dermal atrophy [8].

CONCLUSION

Thus as indicated by data in our study, the problem of TCS is already rampant in all parts of our country. With increasing magnitude of problem, quick and urgent steps are required to increase the awareness and for its management.

Statement of human and animal rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

Statement of informed consent

Informed consent was obtained from all patients for being included in the study

REFERENCES

1. Hughes J, Rustin M. Corticosteroids. Clin Dermatol. 1997;15:715-21.
2. Rathi S. Abuse of topical steroid as cosmetic cream: A social background of steroid dermatitis. Indian J Dermatol. 2006;51:154-5.
3. Frumess GM, Lewis HM. Light-sensitive seborrheid. AMA Arch Dermatol. 1957;75:245-8.
4. Leyden JJ, Thew M, Kligman AM. Steroid rosacea. Arch Dermatol. 1974;110:619-22.
5. Zmegac ZJ, Zmegac Z. So-called perioral dermatitis [in Croatian]. Lijec Vjesn. 1976;98:629-38.
6. Ljubojeviae S, Basta-Juzbasiae A, Lipozenèiae J. Steroid dermatitis resembling rosacea: aetiopathogenesis and treatment. J Eur Acad Dermatol Venereol. 2002;16: 121-6.
7. Kligman AM, Leyden JJ. Adverse effects of fluorinated steroids applied to the face. JAMA. 1974;229: 60-2.
8. Goldman D. Tacrolimus ointment for the treatment of steroid-induced rosacea: a preliminary report. J Am Acad Dermatol. 2001;44:995-8.
9. Pabby A, An KP, Laws RA. Combination therapy of tetracycline and tacrolimus resulting in rapid resolution of steroid-induced periocular rosacea. Cutis. 2003;72: 141-2.
10. Bhat YJ, Manzoor S, Qayoom S. Steroid-induced rosacea: a clinical study of 200 patients. Indian J Dermatol. 2011;56:30-2.
11. Beltrani VS, Barsanti FA, Bielory L. Effect of glucocorticosteroids on the skin and eye. Immunol Allergy Clin North Am. 2005;25:557-80.
12. Vati RR, Ali F, Teuber S, Chanc C, Gershwin ME. Hypersenstivity Reactions to Corticosteroids. Clin Rev Allergy Immunol. 2014;47:26-37.
13. Liu ZH, Du XH. Quality of life in patients with steroid dermatitis before and after treatment. J Eur Acad Dermatol Venereol. 2008;22:663-9.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Brief Report

Burmese thanaka powder and benedict's reagent to struggle the liaison dangereuse: inverse psoriasis plus intertrigo

Lorenzo Martini[1], Alessandro Valle[2]

[1]University of Siena, Department of Pharmaceutical Biotechnologies, Via A.Moro 2, 53100 Siena, Italy, [2]Morcare Ltd,Via S. Giovanni, 5, Monteu Da Po. Turin, Italy

Corresponding author: Lorenzo Martini, M.Sc., E-mail: martinil163@libero.it

ABSTRACT

It has been possible for me to carry out this case report because I decided to make experimentations directly on my body, and specifically applying phytotherapeutic agent, that might be reputed biological and natural at all, and chemical reagents directly onto my groin affected by a severe inverse psoriasis with a well detectable dermatological complication, like the bacterial intertrigo. The importance of the results resides, in my view, in the fact that I made this experimentations in corpore vili (and I mean on my own corpse) during a special period of the year, the so called Moult of mammals, when usually psoriasis and each of every dermatosis and dermatitis which accompanies the psoriatic manifestation, tend to flourish.

Key words: Limonia Acidissima, Feronia Elephantum, Psoralens, Inverse psoriasis

INTRODUCTION

Aims of this brief report is to demonstrate how mild and controlled oxidation reactions, besides the photo-oxidation reactions of psoralens, well known since 1950 [1-7] can evoke the ring opening of various psoralens and angelicin precursors, in order to evoke the formation of psoralen-DNA-thymine-monoadducts, useful to treat psoriasis.

Psoralens are a class of tricyclic aromatic heterocycles that have been used as chemical probes to study DNA and RNA structures. On exposure to UV light, psoralens form chemical cross-links to the DNA bases in three steps [8-10]. The first step is intercalation of psoralen between two adjacent base pairs. The second step is formation of a monoadduct, i.e., one psoralen photoreacts to one strand of DNA. The third step is the cross-linking of the same psoralen to the other strand of DNA.

Many researchers have confirmed that the formation of monoadduct psoralen-DNA-thymine (to yield the cellular apoptosis) can be the sole responsible of the clinical attempt to cure psoriasis, where the theory is accepted, that psoriasis may have the capacity to develop when the immune system mistakes a normal skin cell for a pathogen, and sending out faulty signals that may cause overproduction of new skin cells, so that the disease could be considered as a neoplastic manifestation at all.

Besides the light-induced cross-linking of double-stranded nucleic acids by psoralens, excellent reviews have appeared, some in recent years, which summarize the chemistry of Psoralen 5-Methoxy Psoralen 8-Methoxy Psoralen 5,8-Dimethoxy Psoralen. Some of these reviews have adequately covered methods of degradation reactions of psoralens beyond the classical and well ascertained photochemical reactions.

How to cite this article: Martini L, Valle A. Burmese thanaka powder and benedict's reagent to struggle the liaison dangereuse: inverse psoriasis plus intertrigo. Our Dermatol Online. 2015;6(4):411-414.

Submission: 20.02.2015; **Acceptance:** 10.09.2015

DOI:10.7241/ourd.20154.110

The methods used to degrade psoralens (primarily in order to determine their structure) have been well described, particularly in papers by Spath and his coworkers [11,12], therefore, only a brief description of three degradation reactions is more than sufficient to understand the problem. Furan-2,3-dicarboxylic acid is obtained when either psoralen or angelicin derivatives are oxidized with alkaline peroxide. Moreover alkoxy furocoumarins with an acetic acid-sulfuric acid mixture may give raise to the primitive phenol and alcohol, after the ring has been opened. Finally the lactone ring can be opened in strong sodium hydroxide solution and the resulting phenolic compound is methylated.

So I have thought the best way for me was to try to obtain the ring opening of the psoralens' precursors contained in Thanaka powder, by the aids of chemical reagents and thus I decided to try to do it onto the biological surface itself (in this case, my epidermis affected by prosiasis) by the use of an oxidizing reagent, where the constituents are all admitted in INCI.

Therefore I made up my mind to employ the Bénédict's solvent to open the ring of the structures of marmin and marmesin, which are the precursors of psoralens, as aforesaid and which go to constitute the chemical composition of leaves and bark of Limonia acidissima.

One liter of Benedict's solution contains 173 grams sodium citrate, 100 grams sodium carbonate, and 17.3 grams cupric sulphate pentahydrate, so that its official INCI is:

Aqua; Sodium Citrate; Sodium Carbonate; Copper Sulphate

And this phase will be designed as Phase B to add to Phase A, that is Thanaka powder to loose in water to give a pinkish paste to spread upon the areas of skin affected by psoriasis.

Stoiechometrically speaking there is not interaction between citric acid (contained in the dried pulp of the fruits) and the components of the Bénédict's reagent.

Besides, since the chemical constituents of Limonia acidissima number excellent bactericidal principles, I have decided to treat my inverse psoriasis accompanied by severe clinical complications like intertrigo, since its microbial component had been well ascertained, and other manifestations.

Actually, the skin in the area of my groin had become more sensitive, so the condition was really a hard challenge to manage and treat. Lesions had caused fissures in the creases of inflamed skin, which were absolutely painful and bleeding. Because of its location, the disease had caused even irritation from rubbing and sweating (the aforementioned bacterial intetrigo), but even yeast and fungal infections were not to be excluded and so much as sexual problems because of discomfort occurred.

MATERIALS AND METHODS

I have Thanaka powder sent directly from Thailand (Thailand-etc-nsn) and used to pour one spoonful of it in few water to yield a pink paste to spread twice a day onto the areas of my groin, at 7.00 a.m. and at 02.00 p.m.

Every morning I used to prepare the new paste to apply routinely.

During the afternoon application, I used to spray even the Bénédict's solution after having spread the paste homogenously.

It is necessary to emphasize that during the 6 weeks of treatment I used to wear dark-cotton, or dark hemp or dark flax pants (preferably black or blue), and never white or clear coloured clothing at all and it is suggestive to stress that I have chosen to undergo this case of self medication by the use of phytotherapeutic agents in the period of the year that corresponds to the second annual Moulting in mammals, that is comprised between September and October.

Ethics

This study was performed on human subjects; thus, all patients were aware of the presence of the study and they were fully informed about the drug and its side-effects.

RESULTS

I have recorded all that I noticed during the 42 days of treatment.

During the first 7 days: I observed a dramatic smoothing of the skin by promoting the shedding of psoriatic scales and immediate whitening of the redness, which had been caused both by the inverse psoriasis and the intetrigo.

During the period comprised between the 8th day and the 21st day: I noticed a decrease of the general inflammation and relief of itching, progressive disappearance of macules (smaller than 2 cm in diameter) and patches (larger than 2 cm).

During the period comprised between the 22nd day and 28th day: I observed the gradual block of the production of cells that are routinely overproduced in psoriasis and the progressive disappearance of skin papules (elevated above the skin surface) and plaques (flat-topped and elevated above the skin surface) and all the other embossments of cutis, even the largest and most important, characterized by the typical symptoms of "calor and rubor".

After the 28th day: the total disappearance of nodules (which were spherical and were below the skin surface) and wheals (papules characterised by severe edema).

After 4-5 months, that is in correspondence of the new first annual Moulting in mammals (april-may) no signs of blossoming of new psoriasis was evident.

DISCUSSION

To tell the truth Talamidaw, the first Burmese King Razadarit's consort before to commit suicide, ordered by her young husband who held to be true she had been infidel to him in 1384 , spread her entire body with a cosmetic paste made up with some and uncertain parts of a tree native to Burma, the Feronia elephantum (alias Limonia acidissima). The majorpart of Historiographers divined this flour was made by the powdered bark and dried fruits of the plant, even if some other scientist reported the paste was made by the roots but absolutely not by the gum resin extracted from Feronia, since the use of this gum was well documented as a cathartical and purgative remedy in pediatrics, and thenceforth the popular credo has been asserting for centuries that this yellowish and pinkish cream serves to cleanse skin and mucosae and to purify body and soul to encounter the deities of Death.

But the legend has it that this yellowish cream has been used by Burmese women for over 2000 years. It has a fragrant scent somewhat similar to sandalwood. The creamy paste is applied to the face in attractive designs, the most common form being a circular patch on each cheek, sometimes made stripey with the fingers known as thanaka bè gya, or patterned in the shape of a leaf, often also highlighting the bridge of the nose with it at the same time. It may be applied from head to toe. Apart from cosmetic beauty, thanaka also gives a cooling sensation and provides protection from sunburn. It is believed to help remove acne and promote smooth skin. It is also an anti-fungal and promotes an indisputed whitening effect on skin spots and chloasmas.

According to Linneus' taxonomy the name of this tree is Limonia acidissima and belongs to the family Rutaceae (Citrus family) [13], confined to India, Pakistan, Sri Lanka and Southeast Asia and the usage of the powder of its bark, dried pulp of fruits and/or leaves is widespread throughout Far East, insofar Hindi call it Kath Bel, Javanese designate it as Kawista, Sanskrit name it Billa, Burmese appeal it Thanaka and Malaysian Belingai.

Nowadays indigenous girls and women, attempting to appear European or North-American ladies (envisaging that all that concerns Western civilization represents a totem of wellness and richness) use to treat their face skin, décolletée and arms with Thanaka and the result is a glittering white skin, so scientists and researchers began to try to discover why the chemical and biological constituents of the powdered bark of Limonia acidissima should favour the bleanching of the epidermis.

The pulp is applied onto bites and stings of deadly insects. It is also protective against skin cancer as it can block UV rays. The fruits and stem bark of *L. acidissima* possess larvicidal and antimicrobial activity [14]. Ahmed *et al.* [15] screened the fruit pulp of *L. acidissima* for anti-inflammatory, antipyretic and analgesic activities . A study by Darsini *et al.* 2013, revealed potent antioxidant activity of the fruit and its ability for being used in food and pharmaceutical applications [16].

The preliminary phytochemical analysis of *Limonia acidissima* plant parts showed the presence of alkaloids,flavonoids, phenols, terpenoids, tannins,fats steroids, saponins, glycosides, gum, mucilage and fixed oils [17].

The unripe fruits contain stigmasterol. Fruit pulp contains large quantity of citric acid and other fruit acids, mucilage and minerals. Citric acid is to be reputed the bleanching agent par excellence. Alkaloids, coumarins, fatty acids and sterols have been detected in the pericarp. It also contains umbelliferone, dictamnine, xanthotoxol, scoparone, xanthotoxin, isopimpinellin, isoimperatorin and marmin.Leaves contain stigmasterol, psoralen, bergapten, orientin,

vitedin, saponarin, tannins and an essential oil. Marmesin, feronolide and feronone have been isolated from the bark.Seeds contain fixed oil, carbohydrates, proteins and amino acids. Roots contain feronia lactone, geranylum belliferone, bargapten, osthol, isopimpinellin, marmesin and marmin.

It is suggestive to stress that marmin and marmesin, which are contained in the bark, the main constituent of Thanaka powder, are the very precursors of psoralens.

Psoralens are anyhow contained in the root at extremely high percentages (admitted that in some Thanaka powders from some Asian country the presence of root powder can be detected though).

After having considered the amazing and disconcerting results I have obtained, I intend to attempt to assert that there would be a natural remedy against inverse psoriasis and bacterial intertrigo, I deem one of the most vexing skin affections because of the implicit social and sexual discomforts.

CONCLUSION

Six weeks of a mere application of a natural paste from Burma and a simplest chemical reagent are more than sufficient to hold off psoriasis, almost for one whole year, since, everybody knows, psoriasis in the Third Millennium, can not be but held off.

And also people that can not be designed as physicians, surgeons or dermatologists, know well by now.

Statement of Human and Animal Rights

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008.

AKNOWLEDGEMENTS

It has been possible to have the thanaka powder, the uncontaminated, pure officinal herb not counterfeit, as too often happens in the world market deriving from Far East, thanks to intervention, experience and scrupulousness of Dr. A. Valle, Morcare Ltd.

REFERENCES

1. Wessely GV, Dinjaski K. Action of light on substances of the type of furo-coumarins. Monatsh. 1934;64:131.
2. Lerner AB, Denton CR, Fitzpatrick TB. Clinical and experimental studies with 8-methoxypsoralen in vitiligo. J Invest Dermat. 1953;20:299.
3. Patzak R, Neugebauer L, Ueber Polarographische Untersuchungen von Coumari-nen II. Monatsh. 1952:83:776.
4. Mukerji B. Psoralea and other indigenous drugs used in leucoderma. J Sci Ind. Research. 1956;15A:1.
5. Fowlks W. The mechanism of the photodynamic effect. This Symposium, 1956:233.
6. Brokke ME, Christensen BE. Psoralene I: Certain reactions of xantho-toxin. J Org Chem. 1958;23:589.
7. Nakabayashi T, Tokoroyama T, Miyazaki H, Isono S. Studies on coumarin derivatives II. Ultraviolet absorption spectra of coumarin derivatives. J Pharm Soc Japan. 1953;73:669.
8. Isaacs ST, She CKJ, Hearst JE, Rappoport H. Synthesis and characterization of new psoralen derivatives with superior photoreactivity with DNA and RNA. Biochem. 1977;16:1064-8.
9. Johnston BH, Johnson MA, Moore CB, Hearst JE. Psoralen-DNA photoreaction: Controlled production of mono- and diadducts with nanosecond ultraviolet laser pulses. Science. 1977;906.
10. Johnston BJ, Kung AH, Moore CB, Hearst JE. Kinetics of formation of deoxyribonucleic acid cross-links by 4'-(aminomethyl)-4,5',8-trimethylpsoralen. Biochem. 1981;20:735.
11. Spath E. Die naturlichen Cumarine. Ber. 1937;70A:83.
12. Spath E, Kuffner F. Die nattirlichen Cumarine und ihre Wirkung auf Fische. Monatsh. 1936;69:75.
13. Allen BM. Malayan Fruits. An introduction to cultivated species. Donald Moore Press Ltd. Singapore, 1967.
14. Rahman AA, Gopalakrishnan G, Ghouse BS, Arumugam S, Himalayan B. Effect of *Feronia limonia* on mosquito larvae. Fitoterepia. 2000;71:553–5.
15. Ahamed SM, Swamy SK, Jayaverra KN, Rao JV, Kumar VS. Anti-inflammatory antipyretic and analgesic activity of methanolic extract of *feronia limonia* fruit pulp. Pharmacologyonline 2008;3:852-7.
16. Darsini DTP, Maheshu V, Vishnupriya M, Nishaa S, Sasikumar JM. Antioxidant potential and amino acid analysis of underutilized tropical fruit *Limonia acidissima* L. Free Radicals and Antioxidants. 2013.
17. Asha T, Ponnamal N. Preliminary studies on phytochemical and antibacterial activity of Limonia acidissima L. Plant part. Anc Sci Life. 2005;25:57-61.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

A generalized case of purpura annularis telangiectoides of Majocchi

Seray Külcü Çakmak[1], Duru Onan[1], Emine Tamer[1], Nuran Allı[1], Ferda Artüz[1], Servet Güreşçi[2]

[1]Dermatology Clinic, Ankara Numune Education and Research Hospital, Ankara, Turkey, [2]Pathology Clinic, Ankara Numune Education and Research Hospital, Ankara, Turkey

Corresponding author: Assoc. Prof. Seray Külcü Çakmak, E-mail: seraycakmak@gmail.com

ABSTRACT

Purpura annularis telangiectoides is a rare form of pigmented purpuric dermatoses which usually presents as annular patches with central clearing on the lower extremities. We present an atypical presentation of purpura annularis telangiectoides with generalized lesions in an elderly woman.

Key words: Pigmented purpuric dermatoses; generalized; Majocchi

INTRODUCTION

Pigmented purpuric dermatoses (PPD) are a group of chronic, progressive vascular disorders which are resistant to treatment [1]. Purpura annularis telangiectoides (PAT) is an uncommon PPD which is also known as Majocchi disease [2,3]. It is usually seen in adolescents and young adults and characterized by non-palpable annular patches on the lower extremities[2]. Herein we present an atypical case of generalized PAT in an elderly woman patient.

CASE REPORT

An 88-year old women presented with a pruritic eruption which had started 5 months ago. She had hypertention and congestive heart failure and had been using acetylsalycilic acid, metoprolol and losartan potassium therapies for 10 years. Her past medical story was otherwise nonspecific. Dermatological examination revealed multiple red-brown annular patches with central clearing on both of the extremities and trunk (Figs 1 and 2). Laboratory examinations including complete blood count, routine biochemistry, coagulation parameters, urinanalysis were normal.

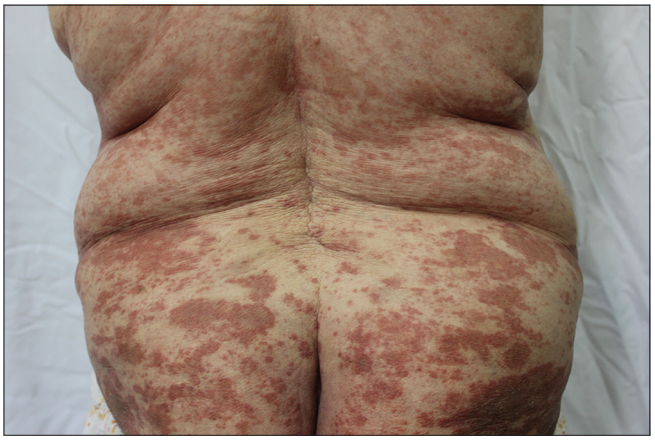


Figure 1: Multiple annular patches the trunk.

Punch biopsy from the lesions showed perivascular lymphocyte infiltration in the papillary dermis and extravasated red blood cells (Fig 3). The patient was diagnosed as PAT and treatment with topical corticosteroid and antihistaminic was started, the lesions showed moderate regression after 15 days.

The patient's informed consent was obtained.
Prior to the study, patient gave written consent to the examination and biopsy after having been informed about the procedure.

How to cite this article: Çakmak SK, Onan D, Tamer E, Allı N, Artüz F, Güre°çi S. Generalized case of purpura annularis telangiectoides of Majocchi. Our Dermatol Online. 2015;6(4):415-417.

Submission: 23.03.2015; **Acceptance:** 10.09.2015

DOI:10.7241/ourd.20153.111

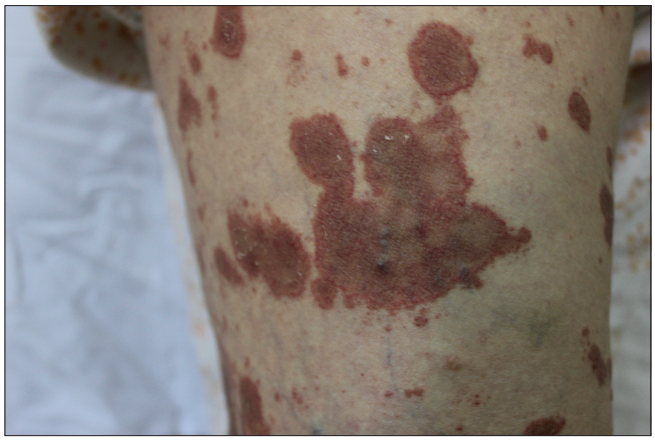


Figure 2: Annular patches with cenral clearing on the lower extremities.

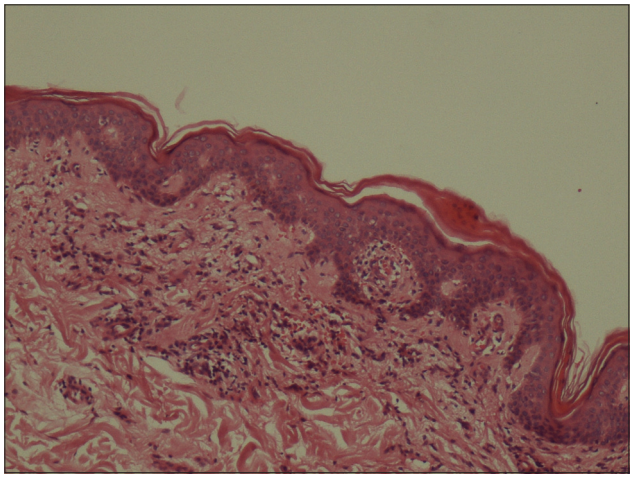


Figure 3: Perivascular lymphocyte infiltration in the papillary dermis and extravasated red blood cells (H&Ex100).

DISCUSSION

PPD are a group of disorders characterized by petechial and pigmentery macules on the lower extremities [1,3]. They are histologically similar and are primarily differentiated on the basis of morphology [4]. PPD include; progressive pigmented purpuric dermatosis, PAT, lichen aureus, pigmented purpuric lichenoid dermatosis and eczematid-like purpura of Doucas and Kapetanakis [1,3].

PAT is a rare form of PPD which was first described in 1896 by Domenico Majocchi [2]. It is usually observed in female adolescents symetrically on the lower extremities [5]. Punctate telangiectatic and purpuric macules progress to annular patches with central clearing and infrequently atrophy [2,4,6]. The lesions are mostly annular but stellate, serpiginous and linear forms have been described [5]. Pruritus is usually not observed [2].

The etiology is unknown, but venous hypertention, exercise, gravitational dependence, capillary fragility, focal infections, contact allergens, medications and alcohol ingestion have been suggested in the pathogenesis [1,3]. Histopathologically superficial papillary dermal capillary dilatation, perivascular lymhocytic infiltration and erytrocyte extravasation are observed [5].

The differential diagnosis of PAT includes leukocytoclastic vasculitis, erythema annulare centrifigum, subacute lupus erythematosus, stasis dermatitis, mycosis fungoides, trombocytopenia and coagulation disorders [5].

Though there is no medication of proven benefit, topical and oral corticosteroids, antihistamines, pentoxyphylline, psoralen-ultraviolet-A and narrowband ultraviolet-B have been used in the treatment [1,3,5].

We presented our case as generalized lesions in an elderly women is a very rare and atypical presentation of PAT. We would like to remind that the differential diagnosis of PAT should be considered in patients with annular erythematous eruptions.

Learning Points

1) Purpura annularis telangiectoides (PAT) can present with generalized lesions
2) The differential diagnosis of PAT should be considered in patients with generalized annular erythematous eruptions
3) PAT can be observed in elderly women

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Sharma L, Gupta S. Clinicoepidemiological study of pigmented purpuric dermatoses. Indian Dermatol Online J. 2012;3:17-20.
2. Hale EK. Purpura annularis telangiectodes of Majocchi. Dermatol Online J. 2003;9:17.
3. Sardana K, Sarkar R, Sehgal VN. Pigmented purpuric dermatoses: an overview. Int J Dermatol. 2004;43:482-8.
4. Hoesly FJ, Huerter CJ, Shehan JM. Purpura annularis telangiectodes of Majocchi: case report and review of the literature. Int J Dermatol. 2009;48:1129-33.
5. Wang A, Shuja F, Chan A, Wasko C. Unilateral purpura annularis

telangiectodes of majocchi in an elderly male: an atypical presentation. Dermatol Online J. 2013;19:19263.
6. Kaplan R, Meehan SA, Leger M. A case of isotretinoin-induced purpura annularis telangiectodes of Majocchi and review of substance-induced pigmented purpuric dermatosis. JAMA Dermatol. 2014;150:182-4.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Beer induced angioedema – A case report

Mrinal Gupta[1], Sarthak Sharma[2], Anish Gupta[3]

[1]Sudhaa Skin Centre, Jammu, India, [2] Department of Radiodiagnosis, Dr Rajendra Prasad Government Medical College, Tanda, H.P, India, [3]Acharya Shri Chander College of Medical Sciences, Jammu, India

Corresponding author: Dr. Mrinal Gupta, E-mail: drmrinalgupta@yahoo.com

ABSTRACT

Beer is a popular alcoholic beverage consumed all over the world. Type I hypersensitivity reactions like urticaria and anaphylaxis due to beer have been reported very infrequently in the literature. We report a case of a 29 year old male who presented with episodes of acute urticaria and angioedema after intake of beer while tolerating other alcoholic beverages. A positive prick test to beer was observed which confirmed the diagnosis of beer induced angioedema. On cessation of beer consumption, no recurrence was observed over a follow-up period of six months.

Key words: Angioedema; Beer; Type I hypersensitivity

INTRODUCTION

Beer is a popular alcoholic beverage consumed all over the world. It mainly contains brewer's yeast, malt, hops and water but wheat, rice, corn and enzymes are added in some special varieties. Barley is the main ingredient of beer which on germination and roasting produces malt which lends beer its richness of flavor and its color. Hops provide the distinctive aroma and bitterness to beer, while yeast *Saccharomyces cerevisiae and S.carlsbergensis* induce fermentation that gives rise to alcohol and carbon dioxide.

In spite of widespread consumption, there have been only a few reports of type I hypersensitivity reactions to beer including urticaria and anaphylaxis which has been attributed to the presence of certain allergens in the barley malt [1-5]. We herein describe the case of a 29 year old male who presented with episodes of angioedema after intake of beer but not with other alcoholic beverages.

CASE REPORT

A 29 year old male presented to us with the history of generalized urticaria and angioedema on two occasions immediately after drinking beer. He was treated with systemic corticosteroids and antihistamines and improved immediately within hours. He also had a history of bronchial asthma and prick tests and specific IgE for pollens of *Compositae* mix were positive. The patient tolerated other alcoholic beverages well and did not develop any symptoms after consuming other alcoholic drinks. Laboratory tests revealed normal blood counts, liver and renal functions but serum IgE level was 440 IU. To confirm our diagnosis of beer induced angioedema, a prick test to beer was performed using histamine phosphate 10mg/ml and normal saline as the positive and negative controls respectively. A wheal with a mean diameter 3mm or more with erythema observed 15 minutes after testing was considered as a positive reaction. A positive prick test to beer was observed in the form of a wheal with erythema measuring 5X5 mm (Fig. 1), thus confirming an immune response against beer. An oral challenge was offered but was declined by the patient. The patient was informed regarding the allergic response to beer and was advised regarding cessation of beer intake. After stopping beer intake, the patient observed no recurrence of symptoms over a follow-up period of six months.

How to cite this article: Gupta M, Sharma S, Gupta A. Beer induced angioedema – A case report. Our Dermatol Online. 2015;6(4):418-419.
Submission: 11.03.2015; **Acceptance:** 10.09.2015
DOI:10.7241/ourd.20153.112

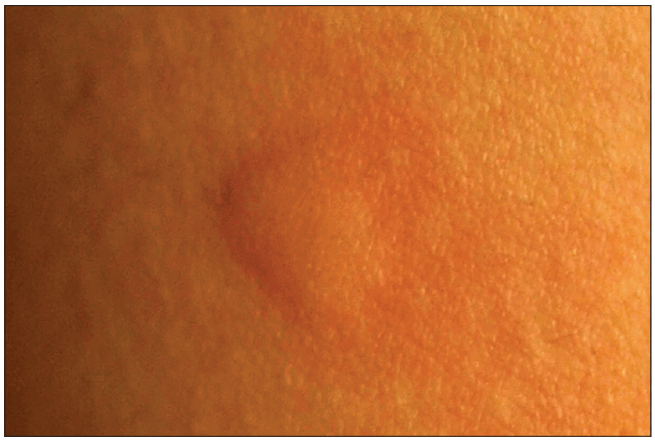


Figure 1: Wheal and flare indicating a positive prick test to beer.

DISCUSSION

Angioedema is a vascular reaction characterized by the development of wheals involving virtually any area of the body. Foods, drugs and physical factors like heat, cold and vibration are the common precipitants of urticaria. Urticaria due to food products occurs due to development of specific IgE antibodies against the specific food allergens. Food induced urticaria is a common entity with reported prevalence rates ranging from 1-2% in children to 6-8% in adults with cow's milk, eggs, peanuts, fish and seafood being the most commonly implicated allergens causing food induced urticaria [6].

Despite its high consumption, allergic reactions to beer have been only rarely reported in the literature. In the reported cases of beer hypersensitivity, barley malt proteins have been identified as the causative allergens in the majority. Beer has been found to contain a large variety of proteins and polypeptides with molecular weights ranging from five to more than 100 kDa which are formed by the proteolytic and chemical modifications of barley proteins occurring during the process of brewing. Curioni *et al* [7] observed that urticaria from beer is an IgE-mediated hypersensitivity reaction induced by a protein component of approximately 10 kDa derived from barley which is unrelated to the major barley 16-kDa allergen responsible for baker's asthma. Generalized urticaria after drinking beer is thought to be due to hypersensitivity to these antigens whereas contact urticaria to beer, without symptoms after drinking, might be caused by differences in end-organ sensitivity.

Hypersensitivity reactions to beer have been seen predominantly among patients with an atopic diathesis. Among the various manifestations reports in the literature, urticaria, especially contact urticaria is the most commonly reported hypersensitivity reaction but a few cases of anaphylaxis have also been reported with beer consumption [1-5]. Van Ketel [2] reported two cases of immediate-type hypersensitivity to beer, one patient developed acute urticaria on entering a bar and anaphylactic shock on drinking beer and the other patient having recurrent episodes of urticaria on consuming beer, with both the patients showing a positive scratch test to malt. Gutgesell *et al* [3] reported the case of a bar waitress who developed hives on her hand upon contact with beer but developed no symptoms on drinking beer. Keller *et al* [4] reported a patient who developed angioedema after drinking beer or eating white bread or cake, the hypersensitivity being attributed to wheat and barley allergens.

In conclusion, hypersensitivity reaction to beer is a rare phenomenon. We report this case of beer induced angioedema to emphasize that beer should also be considered as a potential angioedema inducing agent.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Santucci B, Cristaudo A, Cannistraci C, Curioni A, Furegon L, Giannattasio M. Urticaria from beer in 3 patients. Contact Dermatitis. 1996;34:368.
2. Van Ketel WG. Immediate type allergy to malt in beer. Contact Dermatitis. 1980;6:297–8.
3. Gutgesell C, Fuchs T. Contact urticaria from beer. Contact Dermatitis. 1995;33:436–7.
4. Keller K, Schwanitz HJ. Type I hypersensitivity to beer. Contact Dermatitis. 1994;30:44-5.
5. Figueredo E, Quirce S, del Amo A Cuesta J, Arrieta I, Lahoz C, et al. Beer-induced anaphylaxis: identification of allergens. Allergy. 1999;54:630–4.
6. Hari Sai PV, Anuradha B, Vijayalakshmi VV, Latha SG, Murthy K. Profile of food allergens in urticaria patients in Hyderabad. Indian J Dermatol. 2006;51:111-4.
7. Curioni A, Santucci B, Cristaudo A, Canistraci C, Pietravalle M, Simonato B, et al. Urticaria from beer: an immediate hypersensitivity reaction due to a 10-kDa protein derived from barley. Clin Exp Allergy. 1999;29:407–13.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Bier spots

Ahu Yorulmaz, Seray Kulcu Cakmak, Esra Arı, Ferda Artuz

Ankara Numune Research and Education Hospital, Department of Dermatology, Ankara, Turkey

Corresponding author: Dr. Ahu Yorulmaz, E-mail: ahuyor@gmail.com

ABSTRACT

Also called as physiologic anemic macules, Bier spots are small, hypopigmented irregularly shaped macules against a background of diffuse erythema, which creates an appearance of speckled vascular mottling of the skin. Bier spots most commonly appear on distal portions of the limbs though there are case reports describing diffuse involvement, which also affect trunk and mucous membranes of the patient. Although the exact pathophysiological mechanisms underlying Bier spots still need to be elucidated, Bier spots have been suggested to be a vascular anomaly caused by vasoconstriction of small vessels. In addition, several diseases have been proposed to be associated with Bier spots, including scleroderma renal crisis, cryoglobulinemia, Peutz-Jeghers syndrome, alopecia areata and hypoplasia of the aorta, although it has not been shown whether these associations are casual or coincidental. The clinical presentation of Bier spots is quite typical. These tiny whitish macules easily become prominent when the affected limb is placed in a dependent position and fade away when the limb is raised. Here we report a case of Bier spots in a 32-year-old male patient with characteristical clinical manifestations.

Key words: Bier spots; Etiopathogenesis; Diascopy; Dermoscopy

INTRODUCTION

Bier spots are asymptomatic small, whitish irregularly shaped macules on a background of diffuse erythema, which is tought to be an exaggerated but normal physiological vasoconstrictive response of small vessels to venous congestion [1-7]. It usually affects young adults, more commonly women than men [1,4] and appears to be seen worldwide, in that no racial or ethnic predominance has been reported [4]. Here, we report a 32-year-old male patient with typical clinical presentations of Bier spots.

CASE REPORT

A 32-year-old male patient came to our outpatient clinic with a history of six months of white spots appearing on distal parts of his limbs. His past medical and family history were unremarkable. Systemic examination of the patient was insignificant. Upon dermatological examination on the distal portions of the extremities, we observed multiple whitish macules, several milimeters in size, intermingling with erythrocyanosis of the affected part (Figs 1 – 4). Both flexor and extansor surfaces were affected and the lesions were more pronounced on upper extremities. Lesions became indistinguishable on diascopy (Fig. 5 while dermoscopy provided closer view of eythematous and adjacent blanched white areas (Fig. 6). In addition, when the patient raised his upper extremities above his head, lesions disappeared almost immediately (Fig. 7). Laboratory studies including complete blood count and differential, erythrocyte sedimentation rate, serum chemistry profile, thyroid panel, coagulation profile, urinalysis, C-reactive protein levels, complement levels, immunoglobulin levels and cryoglobulin levels were within normal limits. Anti-nuclear antibodies, antiphospholipid antibodies, rheumatoid factor, proteins C and S were negative. Bilateral upper and lower extremity arterial and venous doppler ultrasonography did not reveal any evidence of pathology. Based on history, clinical, laboratory and ultrasonography findings we made a diagnosis of spontaneous Bier spots and the patient taken under follow-up.

How to cite this article: Yorulmaz A, Cakmak SK, Arı E, Artuz F. Bier Spots. Our Dermatol Online. 2015;6(4):420-423.
Submission: 29.04.2015; **Acceptance:** 10.09.2015
DOI: 10.7241/ourd.20154.113

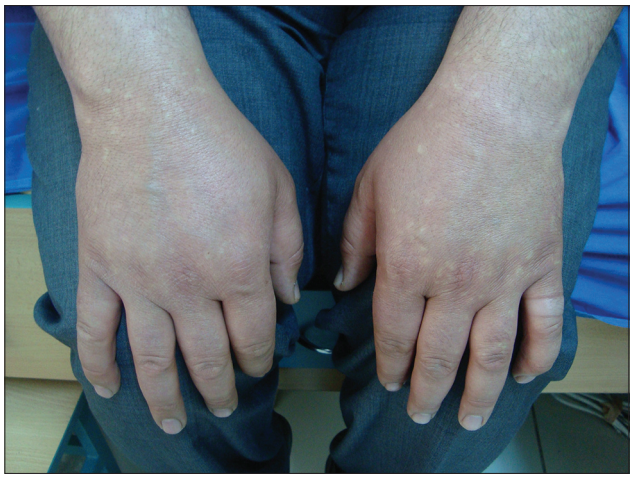


Figure 1: Several scattered white macules within areas of vascular mottling on dorsal surfaces of hands in the dependent position of upper extremities.

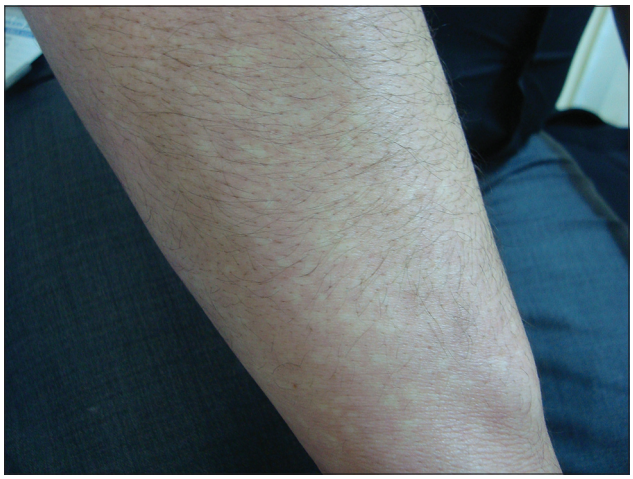


Figure 2: White macules over extansor surface of right forearm

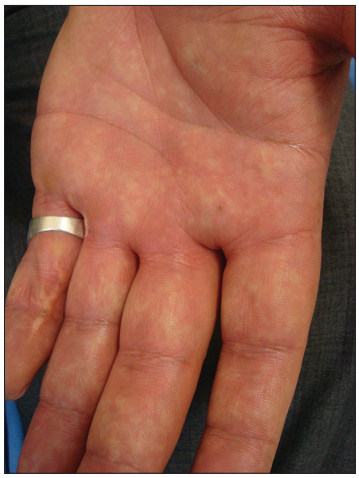


Figure 3: Marked vascular motling on left palmar area.

DISCUSSION

First described in 1898 by Bier [8], Bier spots are asymptomatic pale macules on a background of

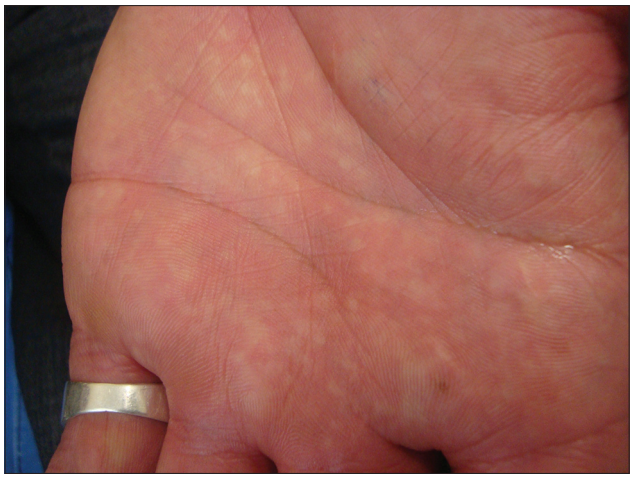


Figure 4: Vascular motling in closer view.

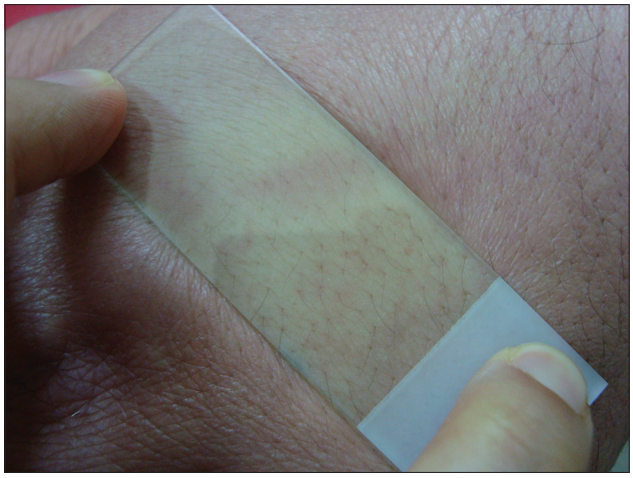


Figure 5: Disappearance of the lesions on diascopy.

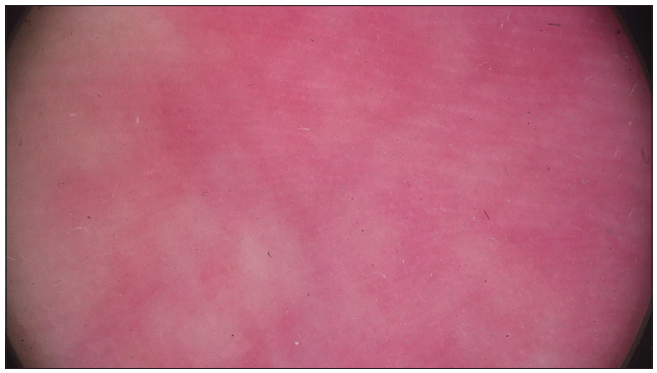


Figure 6: Dermoscopic view (Molemax, x30).

erythema [1,4,7,9-11]. The clinical presentation of Bier spots is unique with typical inducible and reproducible pale macules, several milimeters in size, scattered over dorsal surfaces of extremities in a speckled/mottled appearance. Typically, lesions are visible when the affected limb is put in a dependent position and disappear when the limb is raised. [5-7,9,10,12] Although Bier spots have been regarded to be a

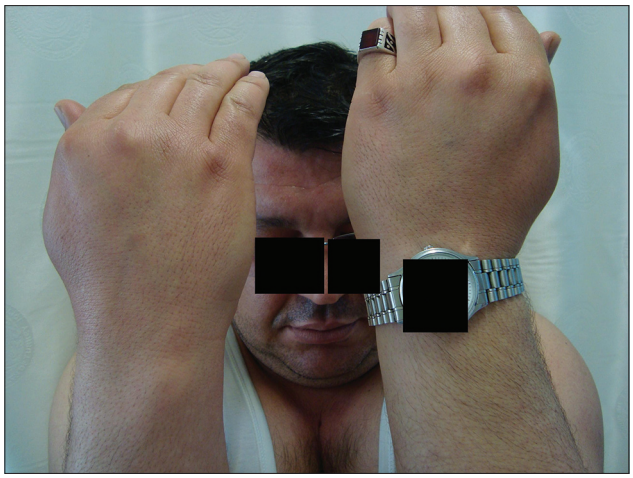


Figure 7: Disappearance of the lesions with limb elevation.

vascular anomaly with vasoconstriction in the paler areas, diascopy, which is an useful and simple technique to evaluate vasodilatation, may be also effective to confirm the diagnosis of Bier spots. Like nevus anemicus, when diascopy is performed, the lesions become indistinguishable from the surrounding skin, since the irregular borders of lesions blanches [4,7].

What are the causes and the reason why Bier spots develop have not been fully elucidated yet. However, a wide range of conditions have been reported to be associated with Bier spots, including pregnancy [13], scleroderma renal crisis [14], cryoglobulinaemia [15], aortic hypoplasia [16], coarctation of aorta [17], lower extremity lymphedema [9], Peutz-Jeghers syndrome, alopecia areata and lichen palnus [7,9]. Moreover, a rare syndrome, Marshall–White syndrome, was described in 1965, which includes Bier spots in conjunction with insomnia and tachycardia [18]. It has been suggested that the pathogenetic mechanism underlying Bier spots is exaggerated vasoconstrictive response within arterioles to tissue hypoxia [2,3,6,9] and causative factors leading to tissue hypoxia have been proposed to be hyperviscosity [15], venous [13] and lymphatic [9] hypertension. However, we think that it is unclear why some individuals develop Bier spots. It has to be clarified whether those aforementioned associations are coincidental or represent a cause and effect relationship or Bier spots are simply develop as a clinical manifestation of exaggerated physiologic vasoconstrictive response.

Here, we report a case of Bier spots in a 32-year-old male patient. We did not find any evidence of underlying conditions, which may cause Bier spots, thus we suggest that our patient represents a case of spontaneous Bier spots. However, as far as we know, Bier spots more commonly appear on the extansor surfaces than the flexor surfaces of the extremities [1,7,9,12] and involvement of palmoplantar area is occasional [4,5,15]. Therefore, since both flexor and extansor surfaces, also palmoplantar areas were involved in our patient, we think that our case is an example of noteworthy presentation of Bier spots. Moreover, to the best of our knowledge, we report herein the first case in the literature describing both the diascopic and dermoscopic examination of Bier spots.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Fan YM, Yang YP, Li W, Li SF. Bier spots: six case reports. J Am Acad Dermatol. 2009;61:e11-2.
2. Collier JG, Dernhorst AC. Bier's spots: evidence that they are mediated by an intravascular vasoconstrictor substance. J Physiol. 1970;209:12e3.
3. Wilkin JK, Marin H. Bier's spots reconsidered: a tale of two sports, with speculation on a humerus vein. J Am Acad Dermatol. 1986;14:411-9.
4. Mahajan VK, Khatri G, Singh R, Chauhan PS, Mehta KS. Bier spots: An uncommon cause of mottled skin. Indian Dermatol Online J. 2015;6:128-9.
5. Heller M. Diffuse Bier spots. Dermatol Online J. 2005;11:2.
6. Tey HL. Spontaneous Bier's spots. Australas J Dermatol. 2008;49:61-2.
7. Tunca M, Caliskan E, Erbil H, Akar A. Bier spots in two children. Pediatr Dermatol. 2011;28:581-3.
8. Bier A. Die Entstehung des Kollateralkreislaufs, II: Der Rückfluss des Blutes. aus ischämischen Korpertheilen. Arch Pathol Anat. 1898;153:306-334.
9. Dean SM, Zirwas M. Bier spots are an under-recognized cutaneous manifestation of lower extremity lymphedema: a case series and brief review of the literature. Ann Vasc Surg. 2014;28:1935.e13-6.
10. Grosshans E. Multiple anemic macules or Bier's spots? Dermatology. 2001;202:272.
11. Khera P, English JC 3rd. Physiologic anemic macules. Cutis. 2008;81:477-8.
12. Liaw FY, Chiang CP. Bier spots. CMAJ. 2013;185:E304.
13. Schoenlaub P, Dupre D, Redon JY, Plantin P. Numerous and large Bier's spots associated with pregnancy. Eur J Dermatol. 1999;9:230e1.
14. Peyrot I, Boulinguez S, Sparsa A, Le Meur Y, Bonnetblanc JM, Bedane C. Bier's white sports associated with scleroderma renal crisis. Clin Exp Dermatol. 2007;32:165e7.
15. Bessis D, Dereure O, Rivire S, Ravi N, Le Quellec A, Guilhou JJ. Diffuse Bier white spots revealing cryoglobulinaemia. Br J Dermatol. 2002;146:921-922.
16. Cabanillas M, Suarez-Amor O, Loureiro M, Ginarte M, Toribio J. Bier's spots in association with hypoplasia of the aorta. Dermatology. 2007;215:166e7.

17. Pearson IC, Holden CA. Delayed presentation of persistent unilateral cutaneous mottling of the arm following coarctation of the aorta. Br J Dermatol. 2003;148:1066e8.
18. White CJ, Marshall W, Kwong LH. Marshall-White syndrome: Evidence of vasomotor conflict in a particularly severe case. J Med Assoc State Ala. 1965;34:249-51.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Kerion Celsi: A report of two cases due to Microsporum gypseum and Trichophyton tonsurans

Edoardo Torres-Guerrero[1], Erick Martínez-Herrera[1], Stefanie Arroyo-Camarena[2], Carlos Porras[3], Roberto Arenas[1]

[1]Mycology section, "Dr. Manuel Gea González" Hospital, Mexico City, Mexico, [2]Dermatology Department "Dr. Manuel Gea González" Hospital, Mexico City, Mexico, [3]Mycology section, Institute of Dermatology and Skin Surgery "Prof. Dr. Fernando A. Cordero C." Guatemala City, Guatemala

Corresponding author: Dr. Edoardo Torres-Guerrero, E-mail: lalotorresg@yahoo.com.mx

ABSTRACT

Tinea capitis is a scalp fungal infection involving the hair. Inflammatory cases are usually caused by zoophilic and geophilic species of the genus *Microsporum* and *Trichophyton*, and are almost always seen in children. The most effective treatments are with Griseofulvin, itraconazole and terbinafine. We report two cases in children 5 and 7 years old, in which *Microsporum gypseum* and *Trichophyton tonsurans* were isolated.

Key words: Kerion Celsi; Dermatophytes; *Microsporum*; *Trichopyton*

INTRODUCTION

Tinea capitis is an infection caused by dermatophyte fungi that affects the scalp and hair; the main pathogens are species of the genera *Microsporum* and *Trichophyton* [1-3]. According to Sarabi and Khachemoune, it can be classified as anthropophilic (*T. rubrum*, *T. tonsurans*, *M. audouinii*, *T. violaceum*), zoophilic (*M.* (*M. gypseum*, *M. fulvum*) [2]. It is almost exclusively in children and rarely occurs after puberty [4].

Inflammatory *tinea capitis*, also known as kerion celsi, is a less common variety, about 13-15% of dermatophytosis of the scalp, usually caused by zoophilic dermatophytes like *M. canis*, *T. mentagrophytes var. mentagrophytes* [2,5], *T. verrucosum*, *T. megninii* (*T. rosaceum*), *T. violaceum* and *T. soudanense*; however, *T. tonsurans* has been documented which may be related to the geographic distribution of microorganisms [4,6]. It is clinically characterized by a solitary and usually very painful well-circumscribed inflammatory lesion, with broken-off hair, and purulent discharge from multiple openings [7].

The inflammation is caused by immunological mechanisms; the lesions may be secondarily infected with bacteria (*S. aureus*) [2] and also, lymphadenopathy [3] may be present.

CASE REPORT

Case 1

A 5 year old boy, presented on the parietal region a 4X4 cm indurated, erythematous scaling plaque with alopecia, and sharp edges; he previously used sulfur soap without improvement. In addition, the patient reported itching and denied fever (Fig. 1).

A superficial scraping of the lesion was performed with a #15 scalpel blade for direct examination with potassium hydroxide and dimethylsulfoxide (KOH-DMSO) where hyaline hyphae and ectothrix hair invasion were visualized.

Mycosel® agar culture (Sabouraud with cycloheximide and chloramphenicol) showed a powdery beige colony,

How to cite this article: Torres-Guerrero E, Martínez-Herrera E, Arroyo-Camarena S, Porras C, Arenas R. Kerion Celsi: A report of two cases due to Microsporum gypseum and Trichophyton tonsurans. Our Dermatol Online. 2015;6(4):424-427.

Submission: 10.04.2015; **Acceptance:** 17.06.2015

DOI:10.7241/ourd.20153.114

suggestive of *Microsporum gypseum* and was confirmed by microscopic examination (Fig. 2).

The patient was treated with griseofulvin (10 mg/kg), with complete resolution, after 45 days. Gastrointestinal symptoms associated to the drug, were the reason to reduce the dose, and increasing the duration of treatment.

Case 2

A 7 year old boy with a lesion on the occipital region of his scalp, characterized by an erythematous plaque of 2.5 cm in diameter, indurated, with alopecia, yellowish scales, with well-defined edges, itchy, and without fever (Fig. 3).

Mycosel® agar showed a yellowish color and radiated colony with powdery appearance, suggestive of *Trichophyton tonsurans var. sulfureum*, confirmed by microscopic examination. The lesion resolved spontaneously within 15 days (Fig. 4).

DISCUSSION

Inflammatory *tinea capitis* or kerion celsi usually occurs in children, starting as a dried form; the inflammatory response is due to immunological mechanisms of the patient [3,8].

The most frequent strains that infect hair and produce inflammatory conditions are zoophilic (*M. canis*, *T. mentagrophytes var. mentagrophytes* and *T. verrucosum*), geophilic (*M. gypseum*), and sometimes, anthropophilic dermatophytes, as *T. tonsurans* [4].

The typical clinical presentation is an inflammatory mass draining pus from multiple openings, usually painful

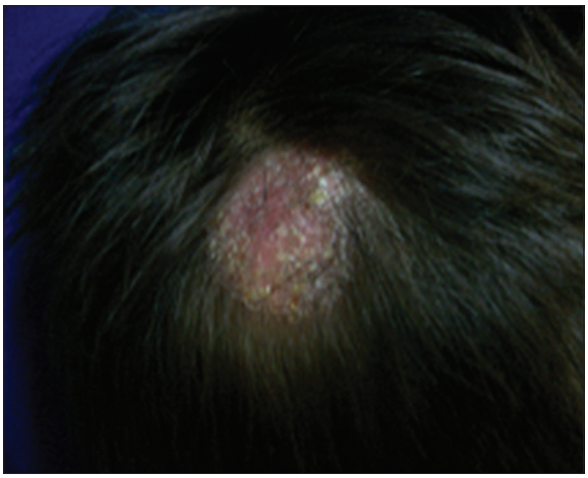


Figure 1: Case 1. Inflammatory *tinea capitis* (kerion celsi).: scaling and pustules.

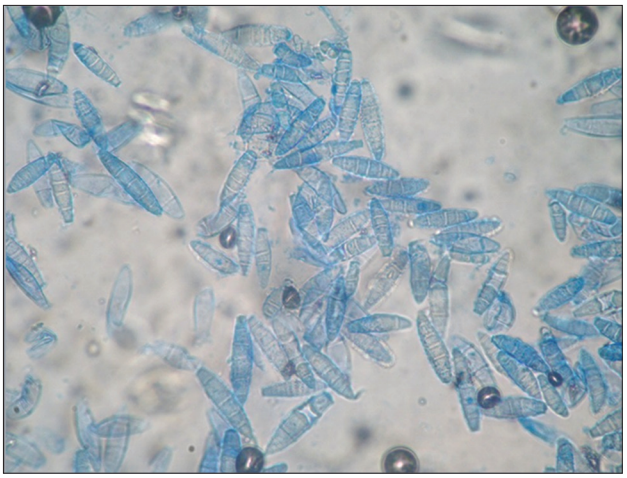


Figure 2: Microscopic aspect of *Microsporum gypseum*: characteristic macroconidia.

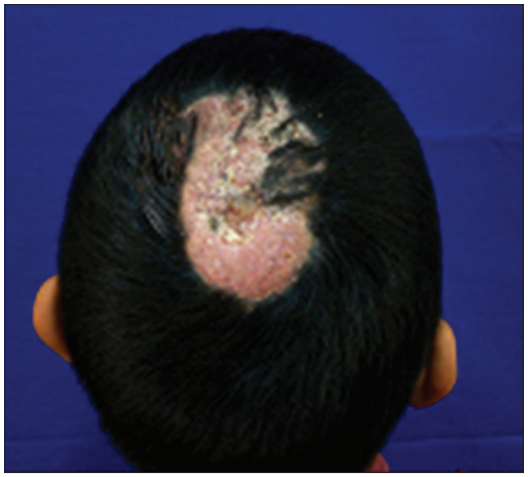


Figure 3: Case 2. Inflammatory *tinea capitis* (kerion celsi). Inflammatory mass with yellowish adherent crusts and pustules.

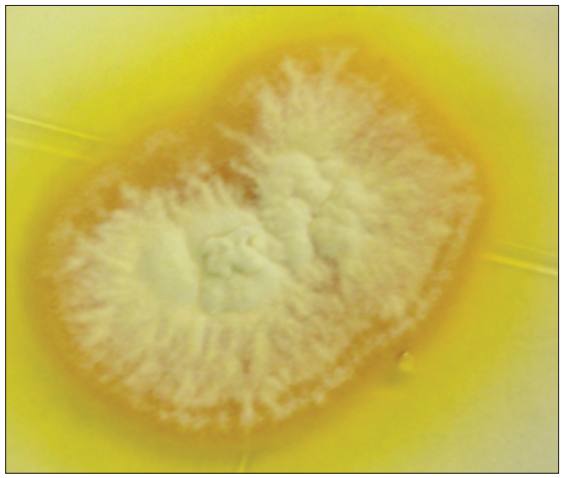


Figure 4: Colony of *Trichophyton tonsurans*.

with pressure; and there is satellite lymphadenopathy with or without fever [1].

Epidemiology may vary depending on the geographical area; the most common etiologic agents in Mexico are *M. canis* (80%), *T. tonsurans* (15%) and sporadically *M. gypseum*, *T. mentagrophytes var. mentagrophytes*, *T. verrucosum* and *T. violaceum* [3]. It is important to note that in the decade of 1960 to 1970 the most prevalent *tinea capitis* in Mexico was trichophytic type (*Trichophyton tonsurans*) and later changed to a predominance of *M. canis* [9]. In the United States the most common etiologic agent is *T. tonsurans*, which overtook *M. canis* in the last 100 years [10].

Other pathogens can be isolated in different regions of the world, as in the Caribbean and Europe (*M. audouinii*, *T. tonsurans*, *T. violaceum* and *M. canis*), in China and Japan (*M. ferrugineum*), Rusia (*T. violaceum*) [3], moreover there may even be variations within the same area, as in a study by Arenas and Torres in which predominance of *T. tonsurans* was observed in rural areas of Dominican Republic, while *M. audouinii* was more common in urban areas of the same country [11]; as in the case of Europe, where in England, *T. tonsurans* is the most common agent for *tinea capitis*, *M. canis* in central and southern Europe and *T. violaceum* in Greece and Belgium [12].

In the first case of this study we isolated *Microsporum gypseum*, known in the literature as a causative agent of inflammatory *tinea capitis*.

M. gypseum is an unusual geophilic pathogen, with a frequency of 0.8% of cases of tinea capitis [13,14]. It occurs in children up to 75% of cases, primarily causing *tinea corporis*, but also *tinea capitis*, *tinea barbae*, *tinea faciei* and onychomycosis. In the scalp it presents as a scaling plaque of alopecia (pseudo-alopecia); pustules, abscesses and golden-yellow crusts are seen in the inflammatory variety [14].

Macroscopic aspect of the culture shows powdery colonies of rapid growth and a light cinnamon color; the reverse may be yellowish. Microscopy shows fusiform and thin-walled macroconidia with less than 6 cells, arranged in groups or clusters. Microconidia are sessile, smooth-walled and club shaped.

In some parts of the world, the most frequently documented agent is *T. mentagrophytes*; in a study by Palacios et al. in Spain, a predominance of *T. mentagrophytes var. granulosum* was observed and they found only one case caused by *M. gypseum* in a child older than 16 years [15]; whereas in a study in Yucatan, Mexico, among 114 cases of *tinea capitis*, both clinical forms (dry and inflammatory) were found in equal proportion, *M. canis* and *T. tonsurans* as the main etiological agents and *T. mentagrophytes*, *M. canis*, *T. tonsurans* and M. *gypseum* for the inflammatory variety [4]. While in a case series study of 19 children with inflammatory *tinea capitis* with biopsy and mycological study, *M. gypseum* was found in 5% of cases [16].

Martinez and Arenas, in a previous study conducted in Guatemala, found the inflammatory form in 36.7% of 60 patients with *tinea capitis*. In the samples collected for this study, the most frequently isolated etiologic agents were *M. canis* (69.8%), followed by *T. rubrum* and *M. gypseum* (13.2% each) [17].

In the second case *Trichophyton tonsurans* was isolated, which morphologically corresponded to the variety *sulfureum*.

There have been reports of *T. tonsurans* in Mexico; in a study of nine cases of inflammatory fungal infections (trichophytic granuloma and kerion), it was found that three cases were caused by *T. tonsurans* and one case by *T. rubrum* [4].

In an outbreak in 5 members of the same family studied in a dermatological center of Mexico City, the inflammatory form was documented in all cases and the causative agent was *T. tonsurans* [18].

T. tonsurans is a common agent of *tinea capitis* and *tinea corporis*. It is the main pathogen of *tinea capitis* in the United States, Canada and England [19,20]. In Mexico it was only identified in 10% of all cases of *tinea capitis* and mainly acquired after contact with an infected person or with fomites [18].

This is an anthropophilic dermatophyte, so animals are never affected, occurring infection by contact from person to person [19].

The macroscopic study shows white-gray colonies, radiated and sometimes with a slight red - brown or yellow pigment on the back. On microscopic examination shows cigar-shaped macroconidia and numerous pyriform microconidia on the sides of the hyphae and in its terminal portion are arranged in a "Cross of Lorraine".

Because of this variation in the frequency of etiologic agents, the importance of regular epidemiological studies is emphasized to present the current status of this clinical entity [9,16].

The first case caused by *M. gypseum* was treated with griseofulvin, which showed good therapeutic response. In the second case, due to *T. tonsurans*, there was no need to prescribe antifungal therapy because of the spontaneous resolution.

Inflammatory dermatophytosis may cause permanent alopecia, therefore should always be treated by a doctor. As important historical antecedent, when no treatments for dermatophytosis were available, cases of scarring alopecia secondary to kerion celsi were relatively frequently.

Currently, the most appropriate treatment is griseofulvin, 10 to 20 mg/kg per day and in resistant cases, up to 30 mg for 8 to 12 weeks. Other successful alternatives are itraconazole and terbinafine; although fluconazole has been used, it is not very effective. Terbinafine can be used to treat infections of *T. tonsurans* and itraconazole for *M. canis* [21]. Prednisone is recommended along with antimycotics to treat the inflammation, and reduce the degree of scarring alopecia [1,16].

CONSENT

The examination of patients is conducted according to the Declaration of Helsinki principles. Written informed consent was obtained from the patient for publication of this case report.

REFERENCES

1. Arenas R. Micología Médica Ilustrada. 4ª ed. Mc Graw Hill, México 2011;61-91.
2. Sarabi K, Khachemoune A. Tinea capitis: A review. Dermatol Nursing. 2007;19:525-9.
3. Bonifaz A. Micología Médica Básica. 3ª ed. México, Mc Graw Hill, 2010;101-119.
4. López–Bárcenas A, Atoche–Diéguez C, Cerón J, Rebollo–Domínguez N, Arenas R. Epidemiologia de la tiña de la cabeza en Yucatán. Estudio de 114 casos. DCMQ. 2009;7:87-90.
5. Asbati M, Bell Smythe A, Cavallera E. Querion de Celso ulcerado por Trichopyton mentagrophytes, var. mentagrophytes. Rev Soc Ven Microbiol. 2002;22:144-6.
6. Isa – Isa R, Arenas R, Isa M. Inflammatory tinea capitis: Kerion, dermatophytic granuloma, and mycetoma. Clin Dermatol. 2010;28:133-136.
7. Bussy RF, Gatti CF, Guardia CP. Fundamentos en Dermatología Clínica. Buenos Aires J. 2011;30-1.
8. Medina D, Del Carmen M, Fernández R, Arenas R, Bonifaz A. Tinea de la cabeza en adultos: Estudio clínico, micológico y epidemiológico de 30 casos en Ciudad de México. Piel.2003;18:403-8.
9. Arenas R. Dermatofitosis en México. Rev Iberoam Micol. 2002;19:63-7.
10. Gupta A. Summerbell R. Tinea Capitis" Med Micol. 2000;28:255-87.
11. Arenas R, Torres E, Amaya M, Rivera ER, Espinal A, Polanco M, et al. Tinea capitis. Emergencia de Microsporum audouinii y Trichophyton tonsurans en la República Dominicana. Actas Dermosifilogr. 2010;101:330-5.
12. Claire F. Changing face of tinea capitis in Europe. Curr Opin Infect Dis. 2009;22:115-8.
13. Zhang R, Ran Y, Dai Y, Zhang H, Lu Y. A case of Kerion celsi caused by Microsporum gypseum in a boy following dermatoplasty for a scalp wound from a road accident. Med Mycol. 2011;49:90-3.
14. Chanussot C, Arenas R. Querón de Celso por Microsporum gypseum en un niño de 5 años. Dermatol Venez. 2010;48:47-9.
15. Del Palacio A, Cuétara María, Valle A, González A, Almondarain I, Ramos M, et al. Cambios epidemiológicos observados en un decenio en las dermatofitosis del hospital universitario "12 de Octubre" de Madrid: nuevas especies emergentes" Rev Iberoam Micol. 1999;16:101-6.
16. Arenas R, Toussaint S, Isa Isa R. Kerion and dermatophytic granuloma. Mycological and histopathological findings in 19 children with inflamatory tinea capitis of the scalp. Int J Dermatol. 2006;45:215-9.
17. Martínez E, De León S, Pérez E, Pacheco A, Rivas E, Borjas C, et al. Tinea capitis. Informe de 60 casos con parasitación pilar y/o agente causal confirmado. DCMQ. 2009;7:98-101.
18. Rodríguez M, Padilla MC, Martínez JA. Tiña inflamatoria de la cabeza por Trichophyton tonsurans. Comunicación de 5 casos dentro de un mismo núcleo familiar. Rev Cent Dermatol Pascua. 2006;15:26-30.
19. Carrillo Silvestri F, Campos González M, Barba Gómez F, Mayorga Rodríguez J. Epidemia familiar por Trichophyton tonsurans. Dermatología Rev Mex. 1998;42:13-5.
20. Mayorga J, Espinoza Gómez R, Villarreal Parra I, García Vargas A. Tiña de la cabeza. Observaciones clínico – micológicas en 30 pacientes. Dermatol Rev Mex. 1999;43:264-7.
21. Elewsky B. Tinea capitis: A current perspective. J Am Acad Dematol. 2000;1:1-20.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Eosinophilic dermatosis: Wells syndrome. A case report

Celeste Valiente Rebull, Tatiana Moreno, Lizza Salgueiro, Gladys Arguello, Valdovinos Gloria, Beatriz Di Martino Ortiz, Oilda Knopfelmacher, Mirtha Rodríguez Masi, Lourdes Bolla de Lezcano

Dermatology Department, Clinicas Hospital, Faculty of Medical Sciencies, National University of Asunción, Asuncion, Paraguay

Corresponding author: Prof. Beatriz Di Martino Ortiz, E-mail: beatrizdimartino@gmail.com

ABSTRACT

Wells syndrome, part of eosinophilic dermatosis, is characterized by infiltrated plaques with edema and erythema, which resolve without scarring and are accompanied by peripheral blood eosinophilia. Familial cases have been described.We report a case of this group of rare dermatosis, pathological studies are essential for diagnosing as clinically encountered numerous differential diagnoses.

Keywords: Eosinophilic dermatosis; Wells syndrome; Eosinophilic cellulitis

How to cite this article: Rebull CV, Moreno T, Salgueiro L, Arguello G, Gloria V, Di Martino Ortiz B, Knopfelmacher O, Rodríguez Masi M, de Lezcano LB. Eosinophilic Dermatosis: Wells Syndrome. A case report. Our Dermatol Online. 2015;6(4):428-432.

Submission: 11.02.2015; **Acceptance:** 29.08.2015

DOI:10.7241/ourd.20154.115

Our Dermatology Online

Case Report

Dermatosis Eosinofílica: Síndrome de WellsDermatosis Wells. Presentación de caso

Celeste Valiente Rebull, Tatiana Moreno, Lizza Salgueiro, Gladys Arguello, Valdovinos Gloria, Beatriz Di Martino Ortiz, Oilda Knopfelmacher, Mirtha Rodríguez Masi, Lourdes Bolla de Lezcano

Cátedra de Dermatología. Hospital de Clínicas. Facultad de Ciencias Médicas. Universidad Nacional de Asunción. Asunción-Paraguay

Corresponding author: Prof. Beatriz Di Martino Ortiz, E-mail: beatrizdimartino@gmail.com

RESUMEN

El síndrome de Wells, parte de las dermatosis eosinofílicas, está caracterizado por placas infiltradas, con edema y eritema, que se resuelven sin dejar cicatriz y se acompañan de esosinofilia en sangre periférica. Se han descrito casos familiares. Aportamos un caso a este grupo de dermatosis poco frecuentes, donde para el diagnóstico es fundamental la anatomía patológica, ya que clínicamente se plantean numerosos diagnósticos diferenciales.

Palabras Claves: dermatosis eosinofílica, síndrome de Wells, celulitis eosinofílica.

INTRODUCCIÓN

El denominador común de las dermatosis eosinofílicas es la eosinofilia en el examen dermatopatológico, por analogía con las "dermatitis neutrofílicas".

La eosinofilia tisular puede ser idiopática o secundaria, como en parasitosis o reacciones a fármacos, además se puede acompañar o no de eosinofilia periférica [1].

La fisiopatología de las dermatosis eosinofílicas es desconocida. Se tratan de síndromes reaccionales caracterizados por una activación del sistema inmunitario en respuesta a diversos antígenos que inducen la producción de citoquina IL-5, principal factor implicado en el quimiotactismo, la activación y la sobrevida de los eosinófilos [2].

La eosinofilia es considerada arbitrariamente como leve (351 a 1500 cel/mm^3), moderada (>1500 hasta 5000 cel/mm^3) y severa, las que presentan recuentos >5000 cel/mm^3 [2]

En ocasiones no es posible demostrar eosinófilos ni en sangre periférica ni en piel, pero a través de la microscopía electrónica e inmunohistoquímica se demuestra la presencia de las proteínas catiónicas de los gránulos capaces de dañar el tejido, jugando un rol importante en la inflamación del tejido [2].

Se consideran cinco las dermatosis eosinofílicas (DE) mayores: la foliculitis pustulosa eosinofílica (FPE), la celulitis eosinofílica (CE) o síndrome de Wells, la hiperplasia angiolinfoide con eosinofilia (HALE), el granuloma facial (GF) y la úlcera eosinofílica de la mucosa oral (UE) interpretada en el sentido de expresión mucosa de infiltrados eosinofílicos autoinvolutivos de posible topografía diversa [1].

La celulitis eosinofílica fue descripta por Wells en 1971 [3], que llamó dermatitis granulomatosa recurrente con eosinofilia. Esta patología presenta cierta semejanza con la celulitis bacteriana, pero a diferencia de la misma, tiene carácter recidivante e histopatología característica, pero no exclusiva [4].

How to cite this article: Rebull CV, Moreno T, Salgueiro L, Arguello G, Gloria V, Di Martino Ortiz B, Knopfelmacher O, Rodríguez Masi M, de Lezcano LB. Dermatosis Eosinofílica: Síndrome de Wells. Presentación de caso. Our Dermatol Online. 2015;6(4):428-432.

Submission: 11.02.2015; **Acceptance:** 29.08.2015

DOI:10.7241/ourd.20154.115

En 1979 Spiegel acuña el término "Síndrome de Wells" [4].

CASO CLÍNICO

Varón, 58 años, chapista, procedente de área urbana del Paraguay (Sudamérica) con alergia a la penicilina y asma.

Quince meses antes presentó un cuadro de granos pruriginosos en espalda que se extienden a tórax, miembros superiores e inferiores. Se realizó biopsia que informa Prurigo alérgico (presumiblemente a picaduras de artrópodos) y se iniciaron antialérgicos y corticoides tópicos, con buena evolución.

Acude al siguiente año por cuadro de 1 mes de evolución de manchas rojas pruriginosas en piel de tórax y miembros inferiores. Debido a los antecedentes anteriores se inician nuevamente antialérgicos, corticoides tópicos y emolientes. Ante la persistencia de las lesiones, y aparición de otras nuevas, se toman nuevas biopsias de tronco.

Examen Físico:

- Múltiples placas eritematovioláceas infiltradas, entre 3 a 5 cm de diámetros, de bordes regulares, límites netos, distribuidas en miembros superiores, inferiores, rostro, abdomen y tórax.
- Múltiples pápulas eritematosas, de 0,3 a 0,5 cm de diámetros, de bordes regulares, límites netos, distribuidas en tórax (Fig. 1).

Laboratorio: Baciloscopía: negativa. Hemograma: Hb: 14,5 g/dl; Hto: 45%; GB: 6.800/mm^3, N: 58%, Eosinófilos: 6% (408 cél/mm^3); Plaquetas: 236.000 cél/mm^3.

Anatomía Patológica: Infiltración dérmica eosinofílica, con histiocitos y *figuras en llama* (Fig. 2). Ausencia de vasculitis aguda, granulomas o ampolla en esta toma.

Diagnóstico: Síndrome de Wells

Tratamiento y Evolución: Prednisona 50mg/día, antihistamínicos, emolientes, con mejoría del cuadro (Fig. 3). En el siguiente control se inicia Dapsona 50 mg/día, posterior al cual el paciente ya no acude al servicio.

Los pacientes firmaron el consentimiento informado para la realizacion de las biopsias y obtencion de fotos.

COMENTARIOS

El síndrome de Wells o celulitis eosinofílica, se describe principalmente en adultos, siendo la presentación en niños menores de 15 años muy rara y con claro predominio del sexo masculino, pudiendo quedar, a diferencia de los adultos, secuelas como pigmentación reticulada, alopecia cicatricial y parálisis del nervio oculomotor [4]. La mayoría de los

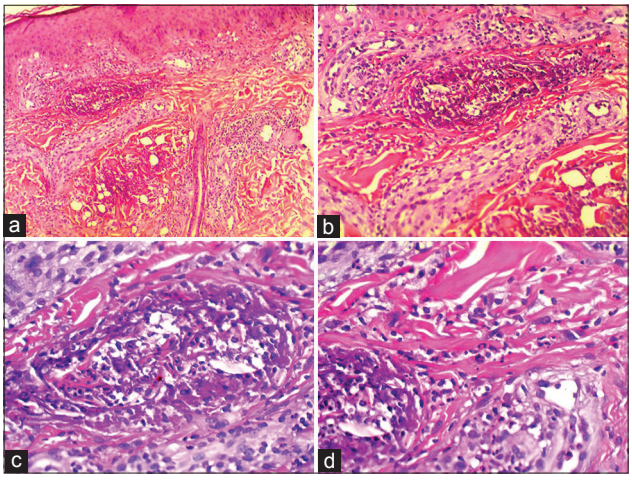


Figure 2: Histopatología. A. En la dermis se observan dos "figuras en llama" (HE 10X). B. A mayor aumento una de ellas (HE 20X). C. La misma está constituida por haces de colágeno degenerado marcadamente eosinófilico rodeado por un infiltrado (HE 40X). D. El infiltrado está compuesto por numerosos eosinófilos e histiocitos (HE 40X).

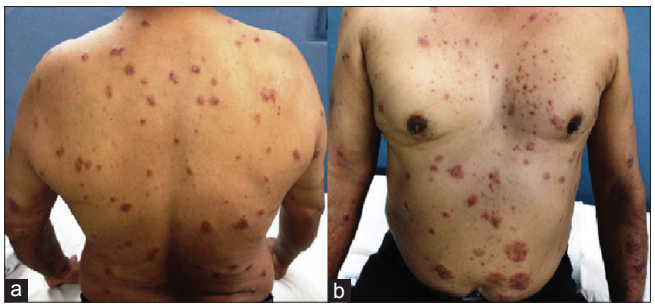


Figure 1: Clínica. Múltiples placas eritematovioláceas infiltradas, distribuidas en miembros superiores, inferiores, abdomen y tórax.

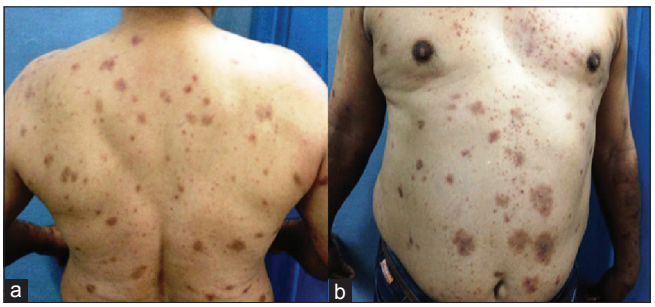


Figure 3: Evolución. Mejoría del cuadro. Las lesions están aplanadas y dejan hiperpigmentación residual.

casos de adultos son autolimitados, sin predilección sexual o racial [5].

El mecanismo patogénico es desconocido [5]. Se señala a una reacción de hipersensibilidad de tipo IV en respuesta a una variedad de estímulos exógenos y endógenos [6].

Se conocen factores desencadenantes de esta reacción de hipersensibilidad como: picaduras de insectos, infecciones virales como herpes virus o bacterianas, erupciones por drogas y las vacunas que contienen timerisol; además han sugerido asociaciones con enfermedades hematológicas, tumores linfoproliferativos y carcinomas [6,7], síndrome hipereosinofílico idiopático, colitis ulcerosa, enfermedad celíaca [7,8]. Muchas drogas han sido relacionadas casualmente al síndrome de Wells, como penicilina, tetraciclina, agentes anticolinérgicos, anestésicos, ácido acetilsalicílico, adalimumab e infliximab [8].

El curso natural puede ser dividido en dos estadios: el primero, que se presenta como placas eritematosas localizadas o difusas, con prurito o ardor. Pueden desarrollar luego edema cutáneo. También podrían presentarse pápulas, nódulos o ampollas. En el segundo estadio, hay involución de lesiones, lo que ocurre en un período de dos a ocho semanas, y podrían quedar secuelas como hiperpigmentación o atrofia residual [6].

El síndrome de Wells es una condición benigna sin manifestaciones sistémicas en la generalidad de los casos, aunque en ocasiones los pacientes pueden describir malestar general, artralgias, rara vez fiebre. Va a recurrir en varias oportunidades en la vida del individuo [9].

Los criterios diagnósticos del Síndrome de Wells son:
1. Máculas eritematosas anulares o edematosas circinadas que pueden evolucionar a placas tipo morfea,
2. La presencia en la histopatología de las "figuras en llama", que no son patognomónicas, y
3. Eosinofilia en sangre periférica que no siempre es constante [8].

Debemos destacar que nuestro paciente cumplía con los 3 criterios, recordando que presentaba una eosinofilia periférica leve.

Para el diagnóstico es fundamental la anatomía patológica, donde en una primera fase o celulítica, podemos observar un edema dérmico con infiltración de leucocitos, predominantemente eosinófilos, que raramente pueden extenderse a la hipodermis o al músculo, sin signos de vasculitis. En la segunda fase, el infiltrado descrito se acompaña de histiocitos y aparecen las características «figuras en llama» compuestas por haces de colágeno degenerado marcadamente eosinófilico rodeado por un infiltrado granulomatoso. Las «figuras en llama» son en realidad un depósito de la proteína básica mayor del eosinófilo en las fibras de colágeno. En la tercera fase o de resolución se produce una desaparición gradual de los eosinófilos, con la presencia aún de histiocitos y células gigantes alrededor de las figuras en llama formando microgranulomas [2]. No suele haber vasculitis [5,8], pero la aparición sucesiva de vasculitis, síndrome de Wells, y síndrome de Sweet se ha informado [8]. Las figuras en llama también se observan en: penfigoide, prurigo, eczema, picaduras de insectos, e infestaciones por parásitos y dermatofitos [3], herpes gestationis [5], en el síndrome de Churg-Strauss [6], mucinosis folicular [6].

En la analítica nos ayuda la eosinofilia, aunque está presente solo en el 50% de los casos. Puede asociarse a un aumento de la VSG y de la inmunoglobulina E [10].

Entre los diagnósticos diferenciales se citan la urticaria, prurigo, síndrome de Sweet [7], urticaria vasculitis, celulitis bacteriana, urticaria por presión, dermatitis de contacto alérgica o irritante, tromboflebitis, lipodermatoesclerosis [8], erisipela, granuloma anular [9], entre otros.

El pronóstico es excelente, y la mayoría de los pacientes se recuperan sin ninguna dificultad [9]. Siempre se deben buscar antecedentes de atopía o urticaria previa y no debemos olvidar realizar un despistaje de procesos oncohematológicos [10].

Los pacientes con eosinofilia idiopática persistente se rotulan dentro del síndrome hipereosinofílico (HES). Pacientes que presentan el espectro HES con hallazgos cutáneos de celulitis eosinofílica y sin enfermedad extracutáneas pueden clasificarse como hipereosinofilia persistente con síndrome de Wells (PHEWS) [11].

Se ha propuesto que la celulitis eosinofílica podría estar en el mismo espectro de enfermedades que presentan trastornos con eosinofilia periférica, como el síndrome hipereosinofílico idiopático (HES) y el síndrome de Churg-Strauss, eosinofilia inducida por fármacos, síndrome de eosinofilia mialgia, foliculitis pustulosa eosinofílica, fascitis eosinofílica, y la hiperplasia angiolinfoide con eosinofilia (enfermedad de Kimura) [12,13].

En cuanto al tratamiento, los corticosteroides sistémicos son el tratamiento más efectivo [14]. De elección es la prednisona a 2mg/kp/día por una semana y luego se inicia el descenso de la misma [6].

Otras terapias son los antihistamínicos anti-H1 (difenilhidralamina 25-50 mg/día), ciclosporina (2,5 a 5 mg/kg/día) y dapsona (50 a 300 mg/día) [5]. Se sugiere el uso de ciclosporina en casos recalcitrantes [6]. Si un agente causal puede ser identificado, puede ser útil tratar la causa subyacente [8].

Los corticosteroides tópicos también demostraron la eficacia, pero se debe considerar en los casos de enfermedades limitadas o en lesiones residuales [6]. El tacrolimus ha sido utilizado con éxito en casos resistentes a esteroides [8].

En conclusión, aunque el síndrome de Wells es una entidad que no se observa todos los días, debe tenerse en cuenta, sobre todo en pacientes que recurren al servicio con lesiones pruriginosas recurrentes. Presentamos el caso debido a la poca frecuencia de esta patología, destacando que a través de la biopsia se pudo llegar al diagnóstico final, debido a que clínicamente planteó numerosos diagnósticos diferenciales. Aportamos este caso de interés y con gran aparatosidad de lesiones clínicas.

BIBLIOGRAFÍA

1. Valdivia-Blondet L. Los eosinófilos y la piel. Dermatol Peru. 2007;17: 83-94.
2. Rodríguez Díaz E, Álvarez Cuesta C, Blanco Barrios S, Galache Osuna C, Requena Caballero C. Dermatosis eosinofílicas I. Actas Dermosifiliogr. 2003;94:65-79.
3. Odia SG, Purschel W, Worret WI, Rakoski J. Hypereosinophilic cellulitis (Well´s syndrome) resembling urticaria. Acta dermatovenerologica A.P.A. 1994;3:193-5.
4. Aparicio S, Torrelo A, Mediero I, Zambrano A. Síndrome de Wells en la infancia. Presentación de un caso y revisión de literatura. Actas Dermosifiliogr. 2000;91:343-8.
5. Hidalgo Parra I, Giusti C, Franco M, Galimberti G, Kowalczuk A, Galimberti R. Síndrome de Wells. Arch. Argent. Dermatol. 2006;56:67-70.
6. Sinno H, Lacroix JP, Lee J, Izadpanah A, Borsuk R, Watters K, et al. Diagnosis and management of eosinophilic cellulitis (Wells' syndrome): A case series and literature review. Can J Plast Surg. 2012;20:91-7.
7. Hassab El-Naby H, El-Khalawany. Multiple erythematous plaques on the trunk and extremities. Gulf J Dermatol Venereol. 2010;17:61-4.
8. Asati D, Arya N, Kudligi C. Well´s syndrome: a case report and review of literature. Int J Biol Med Res. 2012;3:1542-5.
9. Cashin B, Allan N, Kang C. Wells Syndrome. West J Emerg Med. 2010;11:95-6.
10. González Martínez F, Santos Sebastián MM, Navarro Gómez ML, Saavedra Lozano J, Hernández SaMpelayo T. Celulitis eosinofílica: Síndrome de Wells. An Pediatr (Barc). 2009;70:509-11.
11. Powell J, Kaur M, Muc P, Colloby P, Salim A. Hipereosinofilia persistente con síndrome de Wells. Clin Exp Dermatol. 2013;38:40-3.
12. Nguyen N, Ma L. Eosinophilic cellulitis and dermographism. Dermatology Online Journal. 2005;11:7.
13. Herr H, Koh JK. Eosinophilic cellulitis (Wells´Syndrome) successfully treated with low-dose cyclosporine. J Korean Med Sci. 2001;16:664-8.
14. Schwartz RA, Brown J. Chief Editor: Elston D. Aditional Contributors: Butler D, Elenitsas R, Nishikawa T. Wells Syndrome. Updated: May 13, 2014. Disponible en: emedicine.medscape.com/article/1124844.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Hypomelanosis of Ito: Report of two cases

Mohammad Abid Keen

Postgraduate Department of Dermatology, STD & Leprosy Govt. Medical College & Associated SMHS Hospital, Srinagar-Kashmir, India

Corresponding author: Dr. Mohammad Abid Keen, MD, E-mail: keenabid31@gmail.com

ABSTRACT

Hypomelanosis of Ito is a neurocutaneous disorder characterized by hypopigmented whorls, streaks and patches distributed along the lines of Blaschko, often associated with neurological and musculoskeletal abnormalities. We herein report two patients belonging to ethnic Kashmiri origin with this disorder.

Key words: Hypomelanosis of Ito; Neurocutaneous; Lines of blaschko

INTRODUCTION

Ito first described this condition in 1952. Hypomelanosis of Ito is the third most neurocutaneous disorder, after neurofibromatosis and tuberous sclerosis [1]. The characteristic features are the presence of hypopigmented skin lesions arranged in whorls and streaks following the lines of Blaschko. Some believe that Hypomelanosis of Ito is related to autosomal dominant inheritance, others attribute it to chromosomal instability and mosaicism [2]. Chromosomal abnormalities particularly translocation and mosaicism have been reported in approximately 50% cases [3].

CASE REPORTS

Case 1

A 7-month old infant was presented by his parents with hypopigmented lesions over his right upper limb which had been there since birth. These lesions became prominent over time and were not preceded by blistering or any verrucous hyperpigmentation. There was no history of seizure disorder or weakness of limbs. The child was a product of non-consanguineous marriage, born as a full term with normal delivery. There was no history of mental retardation or significant learning disability in any other family member. General physical examination including height, weight and heads circumference were normal. Systemic examination was also non-contributory. Cutaneous examination revealed bizarre hypopigmented linear macules over right arm following the lines of Blaschko (Figure 1). Hair, nails, palms & soles and mucosae were normal. Neurological examination was also normal. There was no musculoskeletal or dental abnormality. Radiological examination of chest, long bones, skull and spine were normal. Ultrasonography of abdomen, 2 D- echocardiography as well as MRI brain revealed no abnormality. IQ testing was not possible because of his age. The child is presently under monthly follow-up.

Case 2

An 8-year old boy presented with hypopigmented lesions over left side of his trunk from second year of his life. These lesions were progressively increasing in size and degree of hypopigmentation. These lesions were asymptomatic and were not preceded by any vesiculobullous, hyperkeratotic or hyperpigmented lesions. There was no history of seizure disorder, neurological deficit or any behavioral disturbance in the patient. The child was a product of non-consanguineous marriage, born at term, second in birth order. There was no history of mental retardation or any significant learning disability in any of the family members. All the developmental milestones

How to cite this article: Abid Keen M. Hypomelanosis of Ito: Report of two cases. Our Dermatol Online. 2015;6(4):433-435.
Submission: 09.01.2015; **Acceptance:** 08.07.2015
DOI: 10.7241/ourd.20154.116

were achieved normally with no developmental delay. General physical examination revealed normal height, weight and head circumference as per his age. Systemic examination was also normal. Dermatological examination revealed whorled hypopigmented macular areas over left side of trunk along the lines of Blaschko (Figure 2). Examination of hair, nails, palms & soles and mucosae was normal. Neurological examination was normal. He had acquired a satisfactory comprehensive and expressive language, with no problems in articulation. The boy evidenced a good level of general awareness, with good vocabulary, calculations and reasoning as per his age. Ophthalmological examination including slit lamp examination and fundoscopy was normal. Musculoskeletal and dental examinations were also normal. Radiological examination of spine, ultrasonographgy of abdomen and echocardiography were all normal. MRI brain as well as ECG revealed no abnormality. IQ testing carried out was also normal. Skin biopsy was not performed as the parents were unwilling for the same. Chromosomal study was not possible in our institute. The child was advised a monthly follow-up.

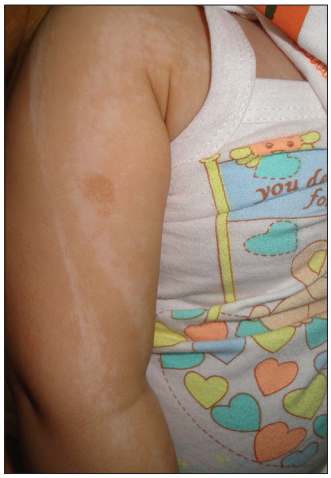


Figure 1: Bizarre hypopigmented linear macules over right arm following the lines of Blaschko.

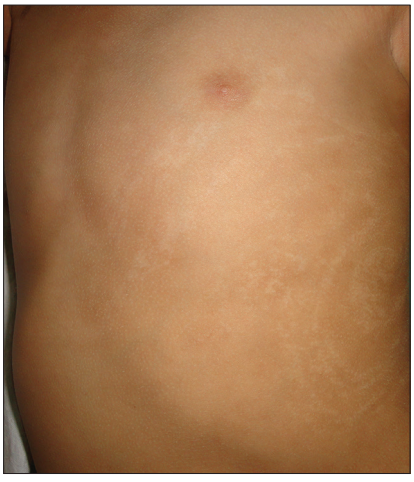


Figure 2: Whorled hypopigmented macular areas over left side of trunk along the lines of Blaschko.

DISCUSSION

In 1952, Minor Ito described a Japanese girl with skin of the upper part of the body looking as "if the normal pigment was brushed off". The depigmented skin lesions were widespread and symmetric, arranged in irregular shapes with "zig zag borders and splash-like spots" on the trunk and in a "linear pattern" down her arms. He defined these lesions as "nevus depigmentosus systematicus bilateralis" [4]. At that time, Ito coined the term "Incontinentia pigmenti acromians" because the pattern of color loss was similar to that of the hyperpigmented changes seen in incontinentia pigmenti of the Bloch-Sulzberger type. Proposed changes in terminology included the terms pigmentary dysplasia, pigmentary mosaicism, pigmentary mosaicism of the Ito type, or hypopigmentation along the lines of Blaschko to reflect the disease pathogenesis or recall the cutaneous patterns [5-7]. Despite these criticisms the term 'hypomelanosis of Ito' is still used.

Hypomelanosis of Ito is a multisystem disorder in which most organs of the body show anomalies in addition to the skin. The main features that define hypomelanosis of ito are the cutaneous anomalies. In many instances, patients may present with skin hypopigmentation following the lines of Blaschko, without any other associated anomaly. The pigmentary lesions are either recognizable at birth or become visible during early childhood. Though most of the cases have been reported from Japan, hypomelanosis of ito has been reported from several other countries [8]. Most reported patients are less than 10 years old and sex ratio favored females by 2.5:1. The characteristic clinical features include unilateral or bilateral areas of hypopigmentation with irregular borders arranged in whorls, or linear patterns along the lines of Blaschko. The hypopoigmented zones in hypomelanosis of ito can be seen in any part of the body: head, neck, face, trunk or extremities [9,10]. Other cutaneous lesions associated with hypomelanosis of Ito include café-au-lait spots, nevus marmorata, angiomatous nevi, nevus of Ota, and Mongolian blue spot.

Nervous system alterations are the most frequently associated extracutaneous anomalies. Anomalies of CNS

may include microcephaly or macrocephaly, cognitive and motor retardation, seizures, ataxia, hyperkinesias and hypotonia [11]. Musculoskeltal disturbances are usually observed in more severe phenotypes. Skeltal defects include short stature, asymmetry with hemihypertrophy or deformities (pectus carinatum or excavatum) and toe anomalies (clinodactyly, polydactyly, syndactyly, brachydactyly) [12]. Ocular alterations are rarely reported and include strabismus, nystagmus, hypertelorism, ptosis, myopia, amblyopic cataracts, corneal opacity, micro-opthalmia, macro-opthalmia, optic nerve hypoplasia and retinal degeneration [13]. Oral manifestation consist of defective dental implantation, partial anodontia, dental hypoplasia or dysplasia, conical teeth and defective enamel. Other systemic anomalies which may be associated include renal disease such as single kidney or ureteral duplication and genitourinary anomalies including cryptorchidism and micropenis [14]. There are a limited number of cases associated with tumors, including cystic teratoma, choroid plexus papilloma, complex mature sacrococcygeal dysembryoma, and dental hamartomas. Rarely, malignancies such as ALL, medulloblastoma and neuroblastoma have been reported.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Ruggieri M, Pavone L. Hypomelanosis of Ito: clinical syndrome or just phenotype? J Child Neurol. 2000;15:635-44.
2. Ruiz-Maldonado R, Toussaint S, Tomayo L, Laterza A, del Castillo V. Hypomelanosis of Ito: diagnostic criteria and report of 41 cases. Pediatr Dermatol. 1992;9:1-10.
3. Lungarotti MS, Martello C, Calabro A, Baldari F, Mariotti G. Hypomelanosis of Ito associated with chromosomal translocation involving XP11. Am J Med Genet. 1991;40:447-8.
4. Ito M. Studies on melanin IX, Incontinentia Pigmenti achromians. A singular case of nevus depigmentosus systematicus bilateralis. Tohoku J Exp Med. 1952;55:57-9.
5. Donnai D. Pigmentary mosaicism. In: Harper J, Oranje A, Prose N, editors. Textbook of Pediatric Dermatology. 2nd ed. Oxford: Blackwell Publishing Ltd; 2006. p. 1509-13.
6. Moss C, Larkins S, Stacey M, Blight A, Farndon PA, Davison EV. Epidermal mosaicism and Blaschko's lines. J Med Genet. 1993;30:752-5.
7. Sybert VP, Pagon RA, Donlan M, Bradley CM. Pigmentary abnormalities and mosaicism for chromosomal aberration: Association with clinical features similar to hypomelanosis of Ito. J Pediatr. 1990;116:581-6.
8. Takematsu H, Sato S, Igarashi M, Seiji M. Incontinentia pigmenti achromians (Ito). Arch Dermatol. 1983;119:391-5.
9. Schwartz MF, Esterly NB, Fretzim DF, Pergament E, Rozenfeid IH. Hypomelanosis of Ito: a neurocutaneous syndrome. J Pediatr. 1997;90:236-24.
10. Pascual-castrovieio I. Hypomelanosis of Ito. Neurologia. 1997;12:300-5.
11. Gómez-Lado C, Eirís-Puñal J, Blanco-Barca O, del Río-Latorre E, Fernández-Redondo V, Castro-Gago M. Hypomelanosis of Ito. A possibly under-diagnosed heterogeneous neurocutaneous syndrome. Rev Neurol. 2004;38:223-8.
12. Trägårdh M, Thomsen CR, Thorninger R, Møller-Madsen B. Hypomelanosis of Ito presenting with pediatric orthopedic issues: a case report. J Med Case Rep. 2014;19:156.
13. Weaver RG Jr, Martin T, Zanolli MD. The ocular changes of incontinentia pigmenti achromians (hypomelanosis of Ito). J Pediatr Ophthalmol Strabismus. 1991;28:160-3.
14. Coward RJ, Risdon RA, Bingham C, Hattersley AT, Woolf AS. Kidney disease in hypomelanosis of Ito. Nephrol Dial Transplant. 2001;16:1267-9.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Pseudoainhum associated with Psoriasis vulgaris

Mrinal Gupta[1], Anish Gupta[2]

[1]Sudhaa Skin Centre, Jammu, India, [2]Acharya Shri Chander College of Medical Sciences, Jammu, India

Corresponding author: Dr. Mrinal Gupta, E-mail: drmrinalgupta@yahoo.com

ABSTRACT

Pseudoainhum is the term applied to constricting bands around the digits or the limb which are either congenital or secondary to another disease. Progression of the constriction bands can lead to irreversible damage and autoamputation of the affected digit. Congenital pseudoainhum usually results due to amniotic bands or adhesions *in utero* while acquired pseudoainhum is usually secondary to trauma, neuropathy, systemic sclerosis or infections like leprosy. Psoriasis is a rare cause of acquired pseudoainhum with only five cases reported till date. We report a case of pseudoainhum secondary to psoriasis vulgaris in a 45 year old male which was successfully treated with topical corticosteroids and systemic methotrexate therapy.

Key words: Pseudoainhum; Psoriasis vulgaris; Autoamputation

INTRODUCTION

Pseudoainhum is the term applied to constricting bands around the digits or the limb which are either congenital or secondary to another disease. It is different from ainhum which is a specific type in which a painful constriction of the fifth toe occurs in adults, with eventual spontaneous amputation, usually seen in young African males. Acquired pseudoainhum is usually secondary to trauma, sensory neuropathy, systemic sclerosis or infections like leprosy [1]. Psoriasis is a rare cause of acquired pseudoainhum with only a few cases reported till date [2-6]. We report a case of pseudoainhum secondary to psoriasis vulgaris in a 45 year old male which was successfully treated with topical corticosteroids and systemic methotrexate therapy.

CASE REPORT

A 45 year old male presented with a three months history of development of generalized reddish scaly lesions which were associated with mild pruritus. After around four weeks, the patient started with pain and swelling of the left index finger which was associated with increased erythema and scaling of the pre-existing lesions. There was no previous history of trauma, fever, sore throat and pain in the left index finger and joints. There was also no past history of any medical illness, drug intake, personal and family history of any skin disorder. On examination, the patient had multiple well defined, erythematous, scaly plaques of psoriasis over the trunk and extensor aspects of elbows, knees and dorsum of hands. The left index finger showed the presence of a large erythematous scaly plaque and a well defined constriction band around the level of distal interphalangeal joint (Fig. 1). The nail of the left index finger was discolored with subungual hyperkeratosis and showed increased curvature while the other nails were normal. Investigations including a hemogram, liver and kidney function parameters and serum electrolytes were within normal limits. Chest X-ray, X-ray of hand and ultrasonography abdomen were normal. A skin biopsy was taken and histopathology was consistent with psoriasis.

The patient was started on emollients, clobetasol ointment 0.05% and 15mg/week methotrexate and there was marked improvement within three weeks of therapy with resolution of the constricting band and decreased erythema and scaling over the psoriasis plaques.

How to cite this article: Gupta M, Gupta A. Pseudoainhum associated with Psoriasis vulgaris. Our Dermatol Online. 2015;6(4):436-437.
Submission: 13.03.2015; **Acceptance:** 20.08.2015
DOI:10.7241/ourd.20153.117

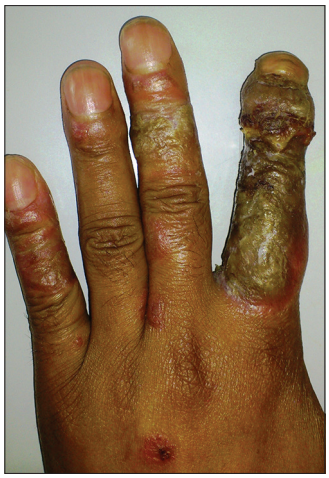


Figure 1: Pseudoainhum of index finger secondary to plaque psoriasis.

DISCUSSION

Pseudoainhum is a rare disease entity which, if left untreated, can lead to irreversible damage and autoamputation of the affected digit or limb. Congenital pseudoainhum usually results due to amniotic bands or adhesions *in utero*, although cases have been reported in Ehlers Danlos syndrome and after amniocentesis [1]. Histology of the band usually reveals broad, finger-like projections of collagen, and coarse elastic bundles which penetrate deep into the subcutaneous fat. Acquired pseudoainhum usually occurs as a result of infections like leprosy and tertiary syphilis, vascular abnormalities like Raynaud's disease, trauma, cold injury, sensory neuropathy, syringomyelia, systemic sclerosis, etc. It may also occur in association with hereditary diseases such as palmoplantar keratodermas like Vohwinkel's disease, pachyonychia congenita, erythropoietic protoporphyria and Olmsted's syndrome. On histology, the bands in acquired pseudoainhum are more superficial and there may be histological features of associated disorders. Surgical management usually in the form of a staged Z-plasty is the preferred treatment modality while in late stages amputation may be required [1,7].

Psoriasis has been described as an uncommon cause of acquired pseudoainhum and till date only five cases of pseudoainhum secondary to psoriasis vulgaris have been reported in the literature. McLaurin [2] reported the acute onset of psoriasis and pseudoainhum development around the middle phalanx of a single digit in a 68 year old female while Kumar *et al* [3] reported acute onset of psoriasis and pseudoainhum development around multiple digits in an adult male. Almond *et al* [4] reported development of pseudoainhum around several digits developing over several months in the presence of long-standing psoriasis. All the three cases required surgical management of constriction bands. Ahn *et al* [5] reported the case of pseudoainhum with psoriasis in a five month old girl which was conservatively managed with topical pimecrolimus and low-dose narrowband UVB phototherapy. Anwar *et al* [6] have reported the development of pseudoainhum over a single digit in an elderly male which was successfully managed with oral acitretin and topical clobetasol propionate ointment.

The pseudoainhum in our patient occurred at the same time that he developed acute exacerbation of psoriasis but rapid initiation of topical and systemic therapy led to complete resolution of the constriction band without causing any irreversible damage. In conclusion, psoriasis is a common disorder which can rarely lead to development of pseudoainhum. It, therefore, becomes necessary to recognize the development and initiate management at the earliest to prevent the progression and autoamputation.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Burrows NP, Lovell CR. Disorder of connective tissue. In: Burns T, Breathnach S, Cox N, Griffiths C, editors. Rook's Textbook of Dermatology, 8th ed. New Jersey: Blackwell Publishing; 2010:46.69-70.
2. McLaurin CL. Psoriasis presenting with pseudoainhum. J Am Acad Dermatol. 1982;7:130-2.
3. Kumar P, Gandhi V. Pseudoainhum in Psoriasis. Indian J Dermatol. 2012;57:238-9.
4. Almond SL, Curley RK, Feldberg L. Pseudoainhum in chronic psoriasis. Br J Dermatol. 2003;149:1064-6.
5. Ahn SJ, Oh SH, Chang SE, Choi JH, Koh JK. A case of infantile psoriasis with pseudoainhum successfully treated with topical pimecrolimus and low-dose narrowband UVB phototherapy. J Eur Acad Dermatol Venereol. 2006;20:1332-4.
6. Anwar MI, Iftikhar N, Hasnain SH, Ishaq BM. Pseudoainhum in Acute Psoriasis. J Coll Physicians Surg Pak. 2012;22:786-8.
7. Brodell RT, Helms SE. Constricting band (Ainhum and Pseudoainhum). In: Wolff K, Goldsmith LA, Katz SI, Gilchrest BA, Paller AS, Leffell DJ, editors. Fitzpatrick's Dermatology in General Medicine, 7th ed. New York: Tata McGraw Hill Publishing; 2008:562-3.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Coexistence of psoriasis and atopic dermatitis

Agnieszka Terlikowska-Brzósko[1], Elwira Paluchowska[1], Witold Owczarek[1], Robert Koktysz[2], Ryszard Galus[3]

[1]Department of Dermatology, Military Institute of Medicine, Szaserów 128 Str., 04-141 Warszawa 44, Poland, [2]Department of Pathology, Military Institute of Medicine, Szaserów 128 Str., 04-141 Warszawa 44, Poland, [3]Department of Histology and Embryology, Center for Biostructure, Medical University of Warsaw, Chalubinskiego 5 Str., 02-004 Warsaw, Poland

Corresponding author: Dr. Ryszard Galus, E-mail: rkgalus@wp.pl

ABSTRACT

Psoriasis and atopic dermatitis (AD) are diseases of still unknown precisely etiology. Concomitance of psoriasis and AD is relatively very rare, but it is constantly under discussion whether these disorders are etiopathologically connected. We report case of 55-year old patient, with a 25 year history of psoriasis, hospitalized in our Department because of exacerbation of atopic dermatitis diagnosed two years ago. We agree with previous reports that due to rare prevalence of concomitance of psoriasis and AD, those diseases are rather mutually exclusive.

Key words: Atopic dermatitis; Coexistence; Psoriasis

INTRODUCTION

Psoriasis and atopic dermatitis are chronic dermatological disorders.

Psoriasis is a recurrent inflammatory skin disease which affects around 2% of the population and is characterized by erythematous papules and plaques covered with silvery white scales. [1]. Atopic dermatitis (AD) is a common skin condition, particularly in children, regarded as one of the 50 most prevalent diseases worldwide [2]. Prevalence of AD in adult patients is around 1-3 % [3]. Patients present various lesions: exudative papules, erythematous plaques, vesicles, erosions with crusts, excoriations and lichenification [1].

Although incidence of psoriasis and AD occurring separately is relatively high, coexistence of both is rare and, thus, not yet sufficiently investigated [4-6].

CASE REPORT

We present a 55-year-old male with over 25 year history of classical clinical picture of psoriasis vulgaris. Course of psoriasis was exacerbated by infections of the upper respiratory tract.

Patient had asthma and confirmed allergy to house dust mite and total IgE substantially elevated. Two years ago, due to the fulfilled 3 of 4 major and 10 out of 23 minor Hanifin and Raika criteria, atopic dermatitis was diagnosed [7].

Patient was hospitalized in our Department of Dermatology because of the exacerbation of atopic dermatitis. On admission has presented two types of lesions: psoriatic ones on the scalp and distal parts of extremities, and typical for AD exudative papules with excoriations and lichenification disseminated on the face, the trunk and extremities. The patient complained of itching of atopic lesions but not of psoriatic ones. Course of skin lesions was specific: while exacerbation of atopic dermatitis was coming, psoriasis had a tendency to remission.

Histopathology taken from the lesion on the left arm revealed features of atopic dermatitis: hypertrophic orthokeratotic stratum corneum; preserved stratum granulosum; spongiosis with vesicle formation in

How to cite this article: Terlikowska-Brzósko A, Paluchowska E, Owczarek W, Koktysz R, Galus R. Coexistence of psoriasis and atopic dermatitis. Our Dermatol Online. 2015;6(4):438-439.

Submission: 01.04.2015; **Acceptance:** 08.06.2015

DOI:10.7241/ourd.20154.118

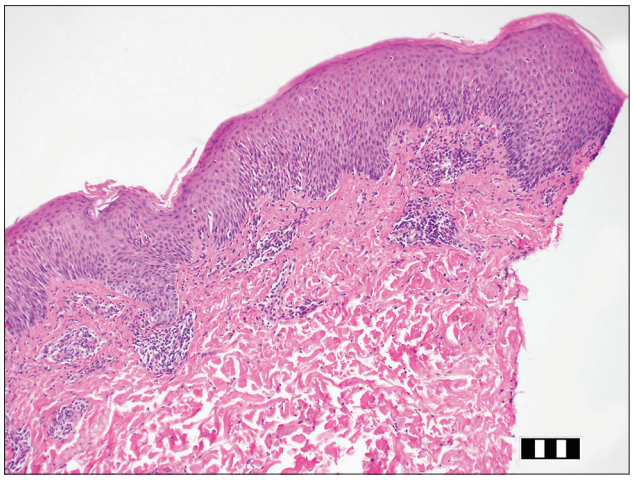


Figure 1: Skin sample collected from clinically confirmed atopic dermatitis in 55-year-old male. Hyper and orthokeratotic stratum corneum; preserved stratum granulosum; spongiosis in acanthotic stratum spinosum; mixed inflammatory infiltrate mostly around vessels in the papillary dermis. Hematoxyline-eosin staining. Scale bar: 100 μm.

acanthotic stratum spinosum; mixed inflammatory infiltration mostly around vessels in the papillary dermis (Fig. 1).

Prior to the study, patient gave written consent to the examination and biopsy after having been informed about the procedure.

DISCUSSION

While new cases of concomitance of psoriasis and AD are still coming, it is constantly under discussion whether these disorders are connected.

Precise mechanisms of AD and psoriasis development are still unclear.

AD demonstrate phenotypical expression of a Th2-driven lymphokine profile and psoriasis is rather Th1-mediated disease [8]. Both diseases appear in genetically predisposed individuals after exposition on same environmental conditions.

In some cases of AD a genetic defect in the filaggrin gene FLG (leading to barrier dysfunction) was proved. Other genetically determined factors involved in pathogenesis of both diseases are under investigation [9,10].

Henseler et al. has statistically proved that relatively very low incidence of concomitance of psoriasis and AD support previous hypothesis by Christophers et al., that both diseases are mutually exclusive [6,8,11].

Presumably, nowadays, we maintain that opinion.

At the end, due to concomitance of AD and psoriasis, we suggest, it would be noteworthy to modify or just wider discuss Eichenfield criteria (for diagnosis of AD), where psoriasis is treated as an exclusionary condition for AD [12].

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Burgdorf WHC, Plewig G, Wolf HH, Landthaler M: Braun–Falco's Dermatology. ed. Springer Medizin Verlag, Heidelberg, 2009.
2. Hay RJ, Johns NE, Williams HC, Bolliger IW, Dellavalle RP, Margolis DJ, et al. The global burden of skin disease in 2010n analysis of the prevalence and impact of skin conditions. J Invest Dermatol. 2014;134:1527–34.
3. Schultz-Larsen F, Hanifin J. Epidemiology of atopic dermatitis. Immunol Allergy Clin North Am. 2002;22:1–24.
4. Kamer B, Rotsztejn H, Kulig A, Raczyńska J, Piotrowicz M, Kulig K, et al. The coexistence of atopic dermatitis and psoriasis in a 12 months-old girl. Pol Merkur Lek. 2005;19:542-4.
5. Beer WE, Smith AE, Kassab JY, Smith PH, Rowland Payne CM. Concomitance of psoriasis and atopic dermatitis. Dermatology. 1992;184:265-70.
6. Dhar S, Kanwar AJ, Ghosh S. Concomitance of psoriasis and atopic dermatitis - a relative phenomenon. Dermatology. 1993;187:76-7.
7. Hanifin JM, Rajka G. Diagnostic features of atopic dermatitis. Acta Dermato-Venereologica. 1980;suppl.92:44-47.
8. Henseler T, Christophers E. Disease concomitance in psoriasis. J Am Acad Dermatol. 1995;32:982-6.
9. Madhok V, Futamura M, Thomas KS, Barbarot S. What's new in atopic eczema? An analysis of systematic reviews published in 2012 and 2013. Part 1. Epidemiology, mechanisms of disease and methodological issues. Clin Exp Dermatol. 2015;40:238-42.
10. Miyagaki T, Sugaya M. Recent advances in atopic dermatitis and psoriasis: Genetic background, barrier function, and therapeutic targets. J Dermatol Sci. 2015;78:89-94.
11. Christophers E, Henseler T. Contrasting disease patterns in psoriasis and atopic dermatitis. Arch Dermatol Res. 1987;279Suppl: S48-51.
12. Eichenfield LF, Hanifin JM, Luger TA, Stevens SR, Pride HB. Consensus conference on pediatric atopic dermatitis. J Am Acad Dermatol. 2003;49:1088-95.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

A case of facial lentiginous lichen planus pigmentosus associated with Hashimoto's thyroiditis and diabetes mellitus

Fadime Kilinc[1], Ayse Akbas[1], Sertac Sener[1], Sibel Orhun Yavuz[2], Ayse Akkus[3], Akin Aktas[3]

[1]Dermatology Clinic, Ataturk Training and Research Hospital, Ankara, Turkey, [2]Pathology Clinic, Ataturk Training and Research Hospital, Ankara, Turkey ,[3]Department of Dermatology, Yildirim Beyazit University Medical Faculty, Ankara, Turkey

Corresponding author: Fadime Kilinc MD, E-mail: fykilinc@yahoo.com

ABSTRACT

Lichen planus pigmentosus (LPP) is an autoimmune, chronic and rare variant of lichen planus of unknown etiology that progresses with pigmentation. The condition is rarely observed concurrently with autoimmune diseases. In this case report, a diabetic male patient with speckled lentiginous lesions on the face, also diagnosed with concurrent autoimmune thyroiditis is presented due to the rarity of the condition and the morphological character of the lesions.

Key words: Autoimmune thyroiditis; Hyperpigmentation; Lentiginous; Lichen

INTRODUCTION

Lichen planus pigmentosus (LPP) is an autoimmune, chronic and rare variant of lichen planus of unknown etiology that progresses with pigmentation [1,2]. The lesions usually occur on the skin sections exposed to the sun or on the flexural regions [3]. The pigmentation is usually diffuse, or may be less commonly reticular, speckled, linear or perifollicular [4]. The condition is rarely observed concurrently with autoimmune diseases. In this case report, a diabetic male patient with speckled lentiginous lesions on the face, also diagnosed with concurrent autoimmune thyroiditis is presented due to the rarity of the condition and the morphological character of the lesions.

CASE REPORT

A 41-year old male patient who has been diabetic for the last six years presented to our outpatient clinic approximately four months ago with facial spots that had suddenly appeared on his face. He had apparent multiple, millimetric, lentigo-like brown pigmented macular lesions on his forehead, temples and cheeks (Figs 1 and 2). His oral mucosa, genital area, scalp and nails were normal. He had no pruritus. He had no history of any cosmetic use. He was on metformin treatment for his diabetes, which is known for its photosensitivity-inducing properties. Based on the pre-diagnoses of pigmented lichen planus, Riehl's melanosis, melasma, and drug-induced hyperpigmentation, a punch biopsy was performed on the pigmented lesion on his face. During the histopathological assessment, the epidermis was observed to be thin with vacuolar degeneration in the basal layer with rare dyskeratotic cells and lymphocytic inflammation in the perifollicular region. Melanophages that have phagocyted melanin have been detected in the papillary dermis (Figs 3 and 4). Based on these findings, the patient was clinically and histopathologically diagnosed with lichen planus pigmentosus. The performed tests also revealed autoimmune thyroiditis (Hashimoto's thyroiditis). His anti-TPO and anti-TG antibodies were very high (Anti-TPO: 600 IU/ml, Anti-TG: 243 IU/ml). His thyroid function tests were normal. And his glucose level was 166 mg/dl. The thyroid ultrasound

How to cite this article: Kilinc F, Akbas A, Sener S, Yavuz SO, Akkus A, Aktas A. A case of facial lentiginous lichen planus pigmentosus associated with Hashimoto's thyroiditis and diabetes mellitus. Our Dermatol Online. 2015;6(4):440-442.

Submission: 30.04.2015; **Acceptance:** 15.06.2015

DOI: 10.7241/ourd.20154.119

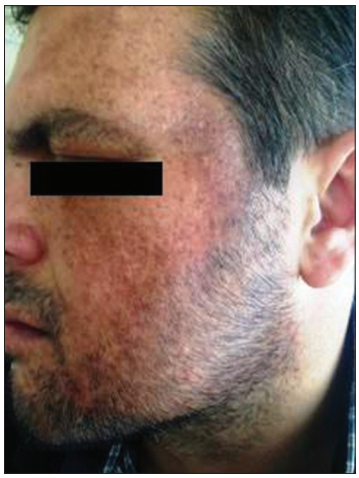


Figure 1: Multiple lentigo-like brown pigmented macular lesions on his forehead, temples and cheeks.

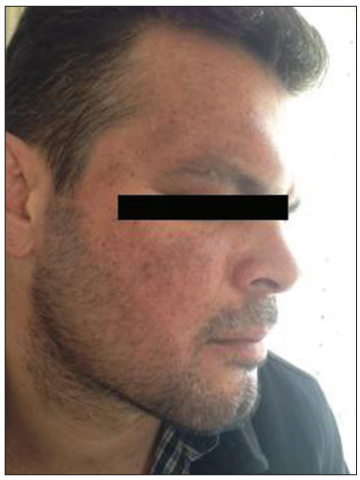


Figure 2: The other side of the face.

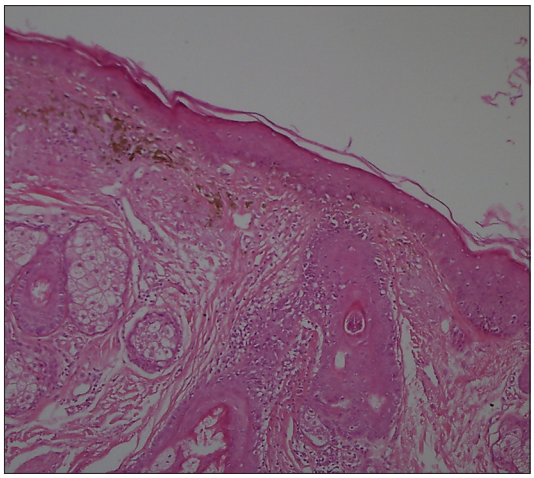


Figure 3: Vacuolar degeneration in the basal layer and perifollicular lymphocytic inflammation, (H&Ex100).

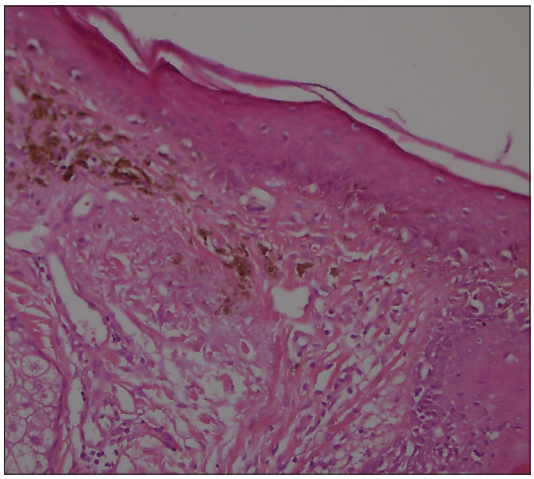


Figure 4: Melanophages in the papillary dermis, (H&Ex200).

revealed thin and thick fibrous bands, and patchy hypoechoic areas. He was recommended to be followed up by the endocrinology department. His hemogram and the other biochemical test results were normal and his hepatitis markers were negative. The patient was recommended a topical treatment with 4% hydroquinone and topical steroid. It was also recommended to change his oral antidiabetic for photosensitivity-inducing properties. The patient was informed about protection from sun.

Prior to the study, patient gave written consent to the examination and biopsy after having been informed about the procedure.

DISCUSSION

LPP is a hyperpigmentation disease first described in Indians by Bhutani et al. [5]. Although its etiology is unknown, exposure to sun, drugs, hepatitis C-virus, internal malignancies, disrupted carbohydrate metabolism, and henna and hair dyes have been suggested as etiological factors [4,6]. T-lymphocyte mediated cytotoxic activity has been blamed in its pathogenesis [6,7].

The mostly affected regions are the face and the neck. Upper extremities and torso may also be involved. Intertriginous lesions are less common [2]. It is generally asymptomatic, while itching may be observed [5]. The lesions are in the form of dark brown macules and papules. Scalp and nails remain unaffected and the oral mucosa is rarely involved. The pigmentation may be diffuse, reticular, speckled, unilaterally linear or perifollicular. The diffuse pattern is the most common pattern and the other forms are very rare [4]. In our patient, the lesions had a speckled pattern. Histopathologically, the epidermis was atrophied with

vacuolar degeneration in the basal layer and lichenoid reaction characterized by infiltration in the dermis in the form of rare lymphocytic bands. Pigment incontinence and melanophages are typical. Also, erythema dyschromicum perstans, fixed drug reaction, postinflammatory hyperpigmentation, and contact dermatitis to the pigment should be considered in the differential diagnosis [2,6].

Hepatitis C, which is more closely associated with oral lichen planus, has been found to be positive in 60,6% of the patients with LPP in a study [4]. Ebrahimi et al. have observed at least one concurrent autoimmune disease in 33 (28%) of the patients with lichen planus [8]. Muzio et al. have detected Hashimoto's thyroiditis in 15 among 105 patients (14.3%) with oral lichen planus and stated that this is a new association, that the thyroid autoantibodies in circulation triggers the organ-specific autoimmune response and leads to lichen planus lesions on the oral mucosa or the skin [9]. Susan et al. have found a significant correlation between autoimmune thyroiditis (15%), alopecia areata (%4), celiac disease (%2) and erosive lichen planus [10]. But, pigmented lichen planus associated with autoimmune diseases has not been reported.

There is no effective treatment for the condition. Potent topical steroids, calcineurin inhibitors, hydroquinone or retinoic acid may be tried. Spontaneous remission may be observed [3,7].

In patients who present with any kind of pigmentation on the face, lichen planus pigmentosus should be among the first diagnoses considered. Also, patients with lichen planus pigmentosus should be scanned for autoimmune diseases. According to our knowledge, our patient is the first case with pigmented lichen planus associated with diabetes and autoimmune thyroiditis.

Autoimmune diseases can affect the resistance to treatment, therefore new researches are necessary about this topic.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Nag F, Ghosh A, Chatterjee G, Choudhary N. Lichen planus pigmentosus: Two atypıcal presentation. Our Dermatol Online. 2013;4:78-9.
2. Jung YJ, Lee YH, Lee SY, Lee WS. A case of lichen planus pigmentosus- inversus in a Korean patient. Ann Dermatol. 2011;23:61-3.
3. Zhang RZ, Zhu WY. One case of linear lichen planus pigmentosus. Open Dermatol J. 2012;6:25-8.
4. Vachiramon V, Suchonwanit P, Thadanipon K. Bilateral linear lichen planus pigmentosus associated with hepatitis C virus infection. Case Rep Dermatol. 2010;2:169-72.
5. Gaertner E, Elstein W. Lichen planus pigmentosus- inversus: case report and review of an unusual entity. Dermatol Online J. 2012;18:11-4.
6. Kashima A, Tajiri A, Yamashita Y, Asada Y, Setoyama M. Two Japanese cases of lichen planus pigmentosus- inversus. Int J Dermatol. 2007;46:740-2.
7. Uyar B, Sivrikoz ON. A case of lichen planus pigmentosus- inversus. Turkderm. 2012;46:160-2.
8. Ebrahimi M, Lundqvist L, Wahlin YB, Nylander E. Mucosal lichen planus, a systemic disease requiring multidisciplinary care: a cross-sectional clinical review from a multidisciplinary perspective. J Low Genit Tract Dis. 2012;16:377-80.
9. Lo Muzio L, Santarelli A, Campisi G, Lacaita M, Favia G. Possible link between Hashimoto's thyroiditis and oral lichen planus: a novel association found. Clin Oral Investig. 2013;17:333-6.
10. Cooper SM, Ali Iaisha, Baldo M, Wojnarowska F. The association of lichen sclerosus and erosive lichen planus of the vulva with autoimmune disease. Arch Dermatol. 2008;144:1432-5.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Rare case report of mesenteric fibromatosis

Radhika Vidyasagar[1], P. Sudarshan[1], Sreedhar Suresh[1], Vidya Bhat[2], M. Subramanya[2]

[1]Department of General Surgery, MVJ Medical College, Bangalore, India, [2]Department of Pathology, MVJ Medical College, Bangalore, India

Corresponding author: Dr. Radhika Vidyasagar, E-mail: radsagar@gmail.com

ABSTRACT

Mesenteric fibromatosis is a part of the clinical-pathologic spectrum of deep fibromatoses. We report this rare case of primary mesenteric tumor that was diagnosed to be a mesenteric fibromatosis on histopathological examination. In majority of patients it may remain asymptomatic and the management of these tumors depends on histopathological examination. Postoperatively, patient was well and subsequent followup showed normal recovery.

Key words: Mesenteric cyst; Fibrzomatosis; Intra abdominal

INTRODUCTION

Mesenteric fibromatosis is a part of the clinical-pathologic spectrum of deep fibromatoses. The deep fibromatoses encompass a group of benign fibroproliferative processes that are locally aggressive and have the capacity to infiltrate or recur but not metastasize.

The deep fibromatoses are classified by anatomic location because they may arise from intra abdominal sites (mesenteric, pelvic, and retroperitoneal fibromatosis), the deep soft tissues of the abdominal wall (abdominal fibromatosis), and deep within extra abdominal soft tissues (extra abdominal fibromatosis).

Mesenteric fibromatosis occurs in a wide age range of patients, 14–75 years of age (mean, 41 years), and has no gender or race predilection.

Thirteen percent of patients with mesenteric fibromatosis have familial adenomatous polyposis (FAP), specifically, the Gardner syndrome variant of FAP. The small bowel mesentery is the most common site of origin of intraabdominal fibromatosis. We report this rare case of primary mesenteric tumor that was diagnosed to be a mesenteric fibromatosis on histopathological examination.

CASE REPORT

A 59-year-old male came to the surgery opd with complaints of pain abdomen since 1 month. On examination, his vitals were normal. On per abdomen examination, there was a solitary swelling measuring 15 x 12 cms, spherical,non tender,freely mobile with a band of resonance heard over the mass. A probable diagnosis of Mesenteric cyst was made.

INVESTIGATIONS

FNAC: Malignant Mesothelioma
Well-Differentiated Adenocarcinoma.

Ultrasound Abdomen and Pelvis

There is a large hyperechoic mass lesion measuring 11.5 x 14.9 x 16.5 cm arising from the Right Iliac Fossa (likely Caecum). There is increased vascularity within the lesion – likely GIST.

NCCT + CECT Abdomen

A well defined, large (measuring 15.6 x 11.3 x 17.5 cm) minimally enhancing, homogenous, solid mass lesion noted in the mesentery with adjacent distal ileal bowel

How to cite this article: Vidyasagar R, Sudarshan P, Suresh S, Bhat V, Subramanya M. Rare case report of mesenteric fibromatosis. Our Dermatol Online. 2015;6(4):443-446.

Submission: 16.02.2015; **Acceptance:** 01.07.2015

DOI: 10.7241/ourd.20154.120

wall thickening (6 mm), mesenteric fat stranding inferiorly, abutting and displacing the adjacent small bowel loops. The lesion is seen in the midline superiorly extending upto origin of SMA, inferiorly up to common iliac bifurcation, anteriorly abutting the anterior abdominal wall. The lesion is supplied by branches of SMA that are seen transversing through out the lesion.

Imp: Large, mesenteric mass lesion as described with adjacent ileal wall thickening – suggestive of GIST, likely to be arising from distal ileum.

PROCEDURE

Patient underwent Exploratory Laparotomy with Intestinal resection and anastomosis under GA on 13/03/2014. A large, solitary mass was present at the root of the mesentery (Fig. 1) and adherent to recto sigmoid colon and small intestine (Fig. 2). Specimen weighed 2.4 kgs (Figs. 3 and 4 and was sent for HPE (Figs. 5 and 6).

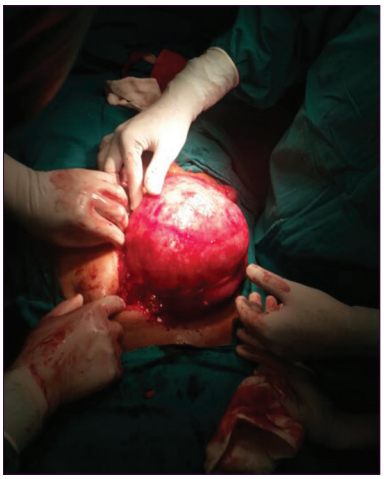


Figure 1: Large abdominal mass present at the root of mesentery.

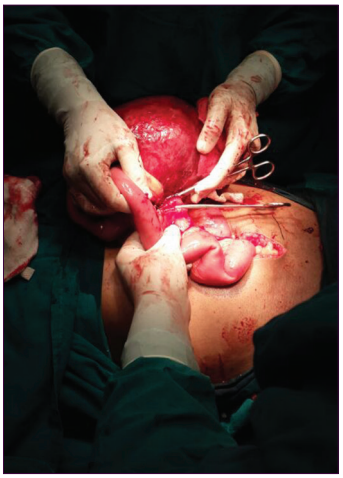


Figure 2: Mass adherent to recto sigmoid colon and the small intestine.

Prior to the study, patient gave written consent to the examination and biopsy after having been informed about the procedure.

DISCUSSION

Mesenteric fibromatosis is a type of fibroblastic proliferation affecting the mesentery that develops usually as a consequence of surgical trauma, but it may

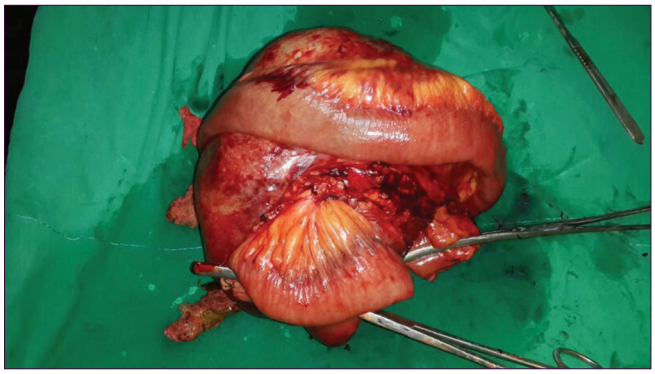


Figure 3: Specimen resected.

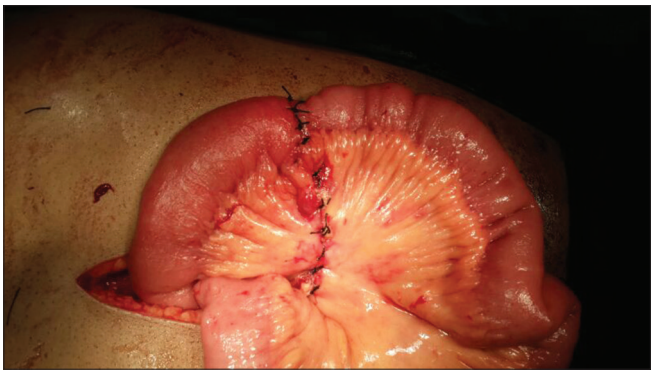


Figure 4: Anastomosis and resection done.

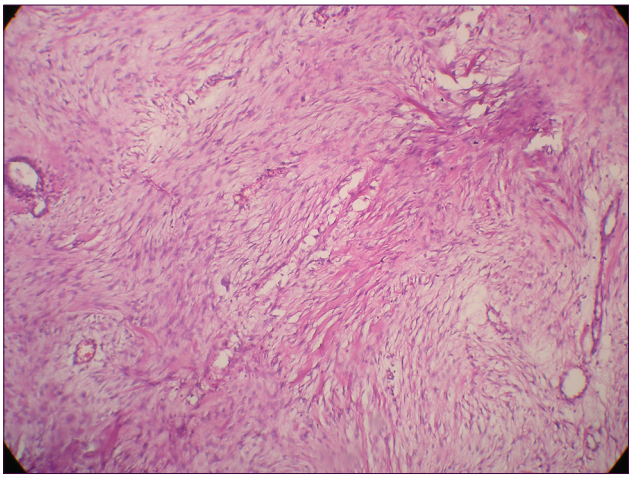


Figure 5: Mesenteric fibromatosis showing cytologically spindle cells in a collagenous stroma.

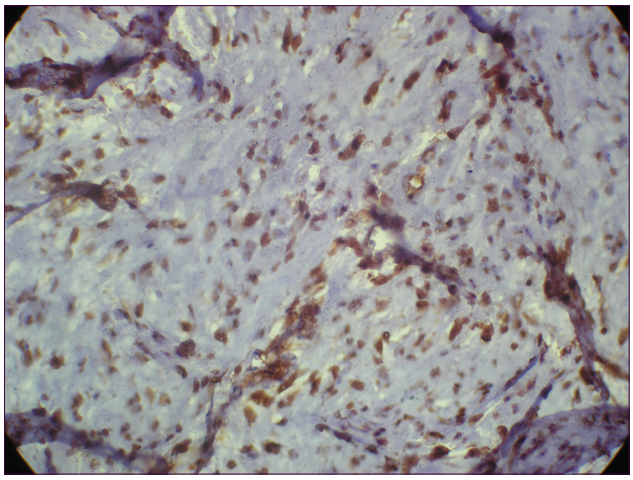


Figure 6: Beta Catenin.

occur spontaneously.It is sufficiently rare for no clear data of incidence and presented features to be known. The majority of patients with mesenteric fibromatosis remain clinically asymptomatic, with little or no focal symptoms until later in their course, at which stage they complain of abdominal pain and discomfort, constipation, vomiting, and organ compression symptoms, such as small bowel obstruction and hydronephrosis.

The etiology is unknown but an endocrine cause is suggested by: (i) the relative prevalence in perinatal women; (ii) tumour regression after the menopause; and (iii) regression with tamoxifen therapy [1].

As there is no classical symptomatology related to mesenteric fibromatosis, the diagnosis is confirmed only after the histological analysis of the tumor. Desmoid tumours are firm masses. On cross section these tumours are grayish and grossly homogenous.

Microscopically, mesenteric fibromatosis is characterized by a spatially homogenous proliferation of wavy spindle cells without atypia, associated with collagen among dilated vessels [2]. The mitotic count is relatively low with no evidence of necrosis and nuclear dedifferentiation.

The diagnosis of mesenteric fibromatosis is based on the microscopic examination and immunostaining. It is noteworthy that a CD117 antigen, expressed commonly in GISTs, can be positive in up to 75% cases of mesenteric fibromatosis. In contrast to GISTs, MF does not express CD34 and S100 protein. Recently, the expression of beta-catenin was revealed in fibromatoses that might prove helpful in the differential diagnosis in doubtful cases [3].

The sonographic features of mesenteric fibromatosis are nonspecific and chiefly dependent on collagen and fibroblast content and intralesional vascularity of the tumor. The results of the treatment might be biased due to the unpredictable course of this disease with some tumours regressing or remaining stable without any treatment.

The management of desmoids should be individualized and multimodal. The surgical resection should be performed only in localized tumours that do not invade the root of the mesentery. Intra-abdominal desmoids can be resected in 53–67% of cases. Fibromatoses are locally aggressive tumours that tend to recur when incompletely resected [4,5].

A high rate of recurrence after the surgical resection results from the incomplete resection, multicentric disease or surgical trauma as a new precipitating factor.

Radiotherapy may be used before surgery in cases of recurrence and inoperable lesions to shrink the tumor and make it operable. Adjuvant radiation therapy reduces recurrence of mesenteric fibromatosis to 20%–40%, compared to 40%–70% with resection only [5]. Recently, a successful therapy of a desmoid tumour resistant to traditional chemotherapeutic regimens was reported with imatinib, a tyrosine kinase inhibitor that is successfully used in advanced gastrointestinal stromal tumours.

CONCLUSION

Mesenteric fibromatosis is a rare tumour that in majority of patients may remain asymptomatic and the management of these tumors depends on histopathological examination.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Kempson RL, Fletcher CD, Evans HL, Hendrickson MR, Sibley RK. Tumors of the Soft Tissues, Armed Forces Institute of Pathology, Washington, DC, USA, 1998.
2. Lotfi AM, Dozois RR, Gordon H, Hruska LS, Weiland LH, Carryer PW, et al, Mesenteric fibromatosis complicating familial adenomatous polyposis: predisposing factors and results of treatment. Int J Colorectal Dis. 1989;4:30-6.

3. Ko SF, Lin JW, Ng SH, Huang CC, Wan YL, Huang HY, et al. Spontaneous isolated mesenteric fibromatosis: sonographic and computed tomographic findings with pathologic correlation. Ultrasound Med Biol. 2006;32:1141–9.
4. Burke AP, Sobin LH, Shekitka KM, Federspiel BH, Helwig EB. Intra-abdominal fibromatosis. a pathologic analysis of 130 tumors with comparison of clinical subgroups. Am J Surg Pathol. 1990;14:335-41.
5. Levy AD, Rimola J, Mehrotra AK, Sobin LH. From the archives of the AFIP: benign fibrous tumors and tumorlike lesions of the mesentery: radiologicpathologic correlation. Radiographics. 2006;26:245–64.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Giant lipoma of the upper back: A case report

Sancar Serbest[1], Uğur Tiftikçi[1], Engin Kesgin[2], Hacı Bayram Tosun[3]

[1]Department of Orthopaedics and Traumatology, Faculty of Medicine, Kırıkkale University, Kırıkkale, Turkey, [2]Department of Orthopaedics and Traumatology, Anamed Private Hospital, Mersin, Turkey, [3]Department of Orthopaedics and Traumatology, Faculty of Medicine, Adıyaman University, Adıyaman, Turkey

Corresponding author: Dr. Sancar Serbest, E-mail: dr.sancarserbest@hotmail.com

ABSTRACT

Lipomas are the most frequent benign tumors of mesenchymal origin. Lipomas may become giant masses, due to usually asymptomatic, painless, slow growing soft tissue tumours. They are more common 5-10 times in males and appearing to avarage age fifth decade of life. Although differantion between lipoma and liposarcoma of low grade malignancy conflict, total surgical excision is adequate treatment. İn this case report, we present 70 years old male with giant back of left shoulder mass as lipoma after surgical excision and histopathological examination.

Key words: Giant lipoma; Shoulder; Surgery

INTRODUCTION

Lipomas originated from the adipose tissue are common, encapsulated and benign neoplasms [1]. They are seen more frequently in the back, shoulder and neck regions. Although their clinical diameters are a few centimeters, they can reach to much larger sizes. Generally, they are seen as a slow-growing, asymptomatic, and painless mass. Even though distribution of men and women is equal, often detected in the fifth decade [2]. To classify lipomas as the giant, they must have 10cm width at least or weight above 1000g [3].

In this study, the lipoma case reached to giant sizes on the localized right shoulder posterior was presented.

CASE REPORT

Seventy years old male patient consulted our polyclinic with painless, slow-growing tumescence complaints which has been existed on the right shoulder for six years. There were also a limitation of shoulder movements and lying back difficulty complaints and no any additional disease or previous trauma history of the patient. During the physical examination, soft viscous mass in the form of two lobes -approximately 25x20x6 cm and 8x10x10 cm- was detected on the dorsal right shoulder (Figs 1a - c). In the radiography, an increase was detected in the soft tissue density in the right shoulder area (Fig. 2). At the contrast-enhanced magnetic resonance imaging (MRI) findings, T1 and T2 weighted images had the same signal with normal adipose tissues. In the fat suppressed images, there were similar kind of suppression with lipomatous tissue (Figs 3a - c). In the MRI of the shoulder joint posterior, there was a view that contains septation and diffuse at the subcutaneous tissue. The mass showed an extension into supraspinotus muscle. At the inferior of the mass, there was also an another lobe which was diffuse, septated, and located at subcutaneous tissue (Fig. 4). Under general anesthesia, it was entered into the incision along the extension of the mass to includes the right shoulder joint of the patient who was lying on the left side. The mass was dissected from subcutaneous tissues, the other part extended into supraspinotus muscle was removed as a mass with the dimensions of 20x18x6 cm. A mass which was 8x8x3 cm and localized as seperate lobe was excised (Fig. 5). During the one year follow-up, no recurrence

How to cite this article: Serbest S, Tiftikçi U, Kesgin E, Tosun HB. Giant lipoma of the upper back: A case report. Our Dermatol Online. 2015;6(4):447-449.
Submission: 07.04.2015; **Acceptance:** 29.05.2015
DOI: 10.7241/ourd.20154.121

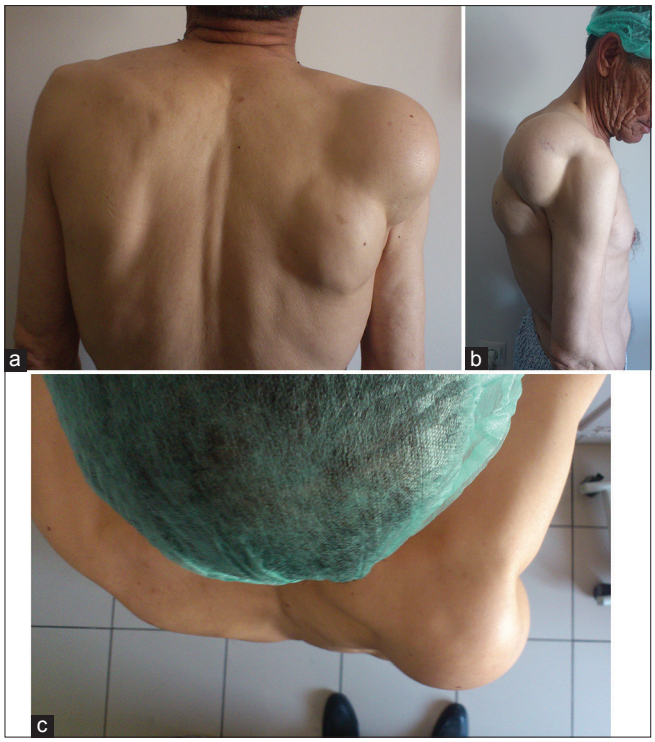


Figure 1: (a, b, c) The physical examination, soft viscous mass in the form of two lobes was detected on the dorsal right shoulder.

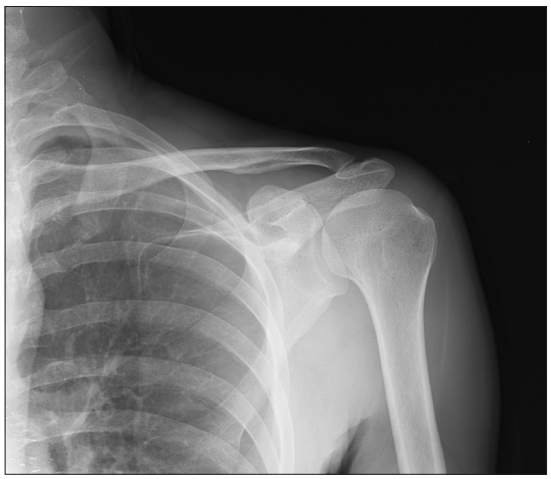


Figure 2: In radiography, there is an increase in soft tissue density at the right shoulder area.

was observed at the patient who was previously reported with the giant lipoma in the histopathological study.

DISCUSSION

Although etiopathenogenesis of lipomas has not been demostrated completely, endocrine, genetic, and traumatic factors are the most accepted causes. Genetically, the possible correlation between lipoma formations and deletion and translocation of 12. chromosome has been informed [4]. The stimulation of secondary inflammatory mediators, fat necrosis, and release of local growth factors are the other factors in charge of the trauma which occurs during the differentiation of mesenchymal precursor cells to adipocytes at the subcutaneus tissues [5].

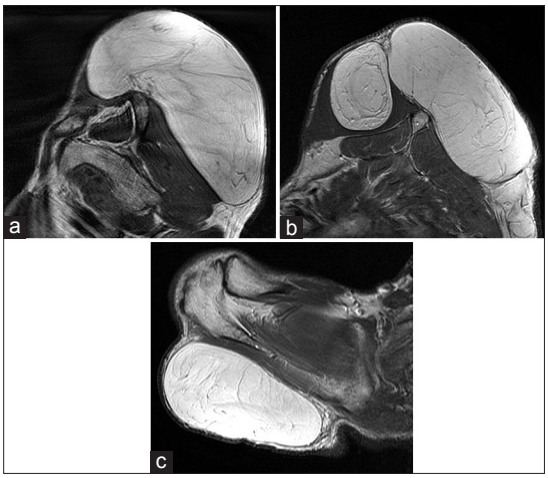


Figure 3: (a, b, c) In contrast-enhanced magnetic resonance imaging, T1 and T2 weighted sections were the same with normal adipose tissue, and there were similar kind of suppression with lipomatous tissue in the fat suppressed images.

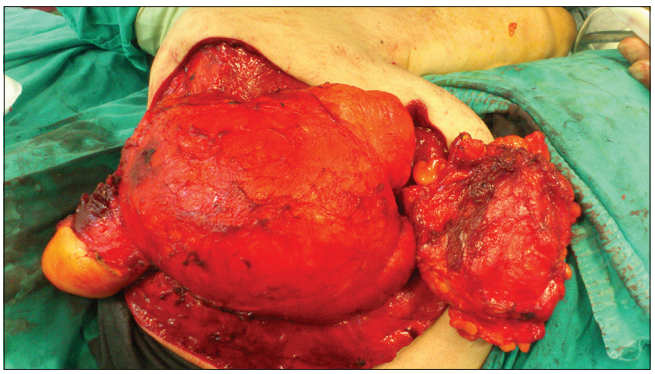


Figure 4: A view which includes diffuse and septation in subcutaneous tissue of posterior shoulder joint.

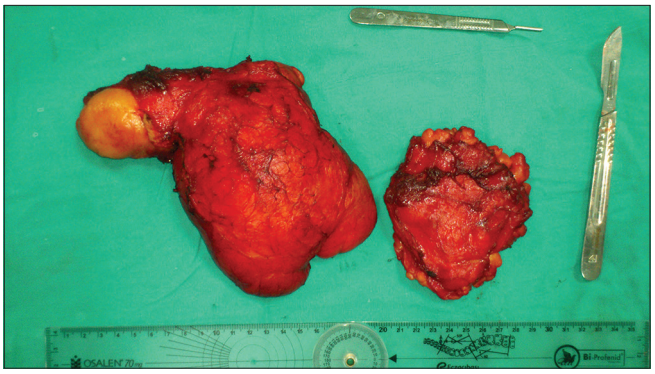


Figure 5: A mass removed as two seperate lobes with sizes of 20x18x6 cm and 8x8x3 cm.

Lipomas are soft viscous, slow-growing, mobile masses which show no sensitivity during the physical examination, create no differences above the skin, may be seen with various sizes depend on their localization and time, and exist for a long time [6]. By reaching large sizes in the body part where they located, they may lead to not only aesthetic but also functional problems. Patients usually consult a doctor because of poor appearence depending on the size of lesion [7].

According to histopathological characteristics, bening lipomatous lesions could be classified as a classic lipoma, a fibrolipoma, an angiolipoma, an infiltrative lipoma, a pleomorpfic lipoma, an intramuscular lipoma and a hibernoma. The study of Üstündağ and Dervişoğlu [8] which covered 843 cases of lipomatous tumor series reported that the malignity was detected only at 44 cases and 75% of benign cases were diagnosed as classic lipomas.

Radiography, ultrasound, computed tomography (CT), and MRI are examination methods used in lipoma diagnosis. Although the radiography is not much sensitive, low density fat opacities can be seen. Ultrasound is easily accessible and the first preferred method because of that it is noninvasive. In the ultrasound, lipomas are generally seen as well circumscribed encapsulated masses. Even though their echogenicities are highly variable, they are iso or hyperechoic according to the muscle. The vascularization is not monitered at Doppler examination. In CT examination, the lipoma is observed as an encapsulated, low density homogen mass. A low density is the definitive diagnostic for the lipoma. MRI imaging is the most frequently used method because of the fact that it has high soft tissue resolution and can show the distribution and depth. Lipomas are not stained with any contrast substance in MRI and CT examinations [9].

The general treatment form of lipomas is a surgical excision. They can be excised due to aesthetic or functional causes. They do not show recurrence after total resection and for this reason additional treatment is not needed [7]. Liposuction is another treatment modality used rarely for giant lipomas [10]. We performed total resection to our case and called the patient for controls. Postoperative consultations at forth and twelfth months were evaluated as normal. To conclude, despite the fact that lipomatous lesions have benign characteristics clinically, defining the histopathological nature of lipomas is absolutely necessary. Difficulty of diagnosis in certain lesions which were evaluated as a simple lipoma by clinicians should be considered.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Kumar V, Abbas AK, Fausto N, Robbins SL, Cotran RS. Robbins and Cotran pathologic basis of disease. 7th edition. Philadelphia, Elsevier Saunders; 2005:XV, p. 1317.
2. Kohler S. Muscle, adipose and cartilage neoplasms. In Bolognia Jl, Jorizzo Jl, Rapini RP. eds. Dermatology, Edinburgh: Mosby; 2003:1883-98.
3. Ghidirim G, Mishin I, Gutsu E, Gagauz I, Danch A, Russu S. Giant submucosal lipoma of the cecum: report of a case and review of literature. Rom J Gastroenterol. 2005;14:393-6.
4. Turc CC, Dalcin P, Boghosian L. Breakpoints in benign lipoma may be at 12q13 or 12q14. Cancer Genet Cytogenetic. 1988:36;131-3.
5. Copcu E, Sivrioglu N. Posttraumatic lipoma: Analsis of 10 cases and explanation of possible mechanisms. Dermatol Surg. 2003:29;215-8.
6. Copcu E, Sivrioğlu N. Posterior cervical giant lipomas. Plast Reconstr Surg. 2005:115;2156-7.
7. Silistreli OK, Durmus EU, Ulusal BG, Oztan Y, Gorgu M. What should be the treatment modality in giant cutaneus lipomas? Review of the literature and report of 4 cases. Br J Plast Surg. 2005:394-8.
8. Üstündağ N, Dervişoglu S. Recently described lipomatous tumours and our 11 year-experience on lipomatous tumours. Cerrahpaşa J Med. 2003;34:119-26.
9. Cree M Gaskin, Clyde A. Helms. Lipomas, lipoma variants and well-differentiated liposarcomas (atypical lipomas): Results of MRI evaluations of 126 consecutive fatty masses. AJR Am J Roentgenol. 2004;182:733-9.
10. Nichter LS, Gupta BR. Liposuction of giant lipomas. Ann Plast Surg. 1990;24:362-5.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

A rare presentation of an ectopic breast tissue in axilla

Radhika Vidyasagar, P. Sudarshan, N. Ravindranath Singh, S. Shivaram

Department of General Surgery, MVJ Medical College, Bangalore, India

Corresponding author: Dr. Radhika Vidyasagar, E-mail: radsagar@gmail.com

ABSTRACT

Accessory breast tissue is rare accounting to less than 1% cases seen in females. It is usually bilateral. We report a case of 24-year-old woman with a lump in the left axilla in view of its rarity and made a differential diagnosis of fibroadenoma, which following the investigations and histopathological report was confirmed as revealed fibroadenoma in the axilla. It should also be considered as a differential diagnosis for all axillary swellings.

Key words: Axilla; Fibroadenoma; Breast

INTRODUCTION

Accessory breast tissue is commonly located above the breast in the axilla. It is uncommon and usually bilateral. Accessory breast tissue may be seen as an enlarging mass in the axilla during pregnancy and persists, as excess tissue in the axilla after lactation is complete. The accessory mammary tissue may be removed surgically if it is large or cosmetically deforming or to prevent enlargement during future pregnancy.

We report a case of 24-year-old woman with a lump in the left axilla in view of its rarity that was histologically identical to the fibro adenoma seen in the breast and also to consider for differential diagnosis of axillary mass.

CASE PRESENTATION

24-year-old woman presented with lump in the axilla since 2 months that increased in size and associated with discomfort. Patient has no family history of breast cancer.

On examination, there was a lump in the left axilla measuring 3 x 2 cms that was firm in consistency, mobile, non-tender. Skin over the swelling was normal. There were no nipple or areola changes seen. Both the breast and right axilla were clinically normal. A provisional differential diagnosis of Axillary lymphadenopathy was made.

Prior to the study, patient gave written consent to the examination and biopsy after having been informed about the procedure.

INVESTIGATIONS

Mammography of the right breast is normal. The visualized left axillary region reveals a focal bulge of homogenous soft tissue density of size approximately 2×2 cm, suggestive of benign SOL in axilla fibroadenoma.

Fine needle aspiration cytology (FNAC) report done outside and in our hospital showed breast tissue with an encapsulated tumor composed of glands and stroma. The glands are predominantly in a pericanalicular pattern, some areas show intracanalicular arrangement. The glands are lined by double-layered epithelium surrounded by proliferative stroma suggestive of fibroadenoma.

TREATMENT

Patient underwent excision biopsy on 5/6/2014. Histopathological examination revealed a globular, glistening soft tissue mass and the cut section showed pale white with slit like spaces (Figs. 1 and 2).

How to cite this article: Vidyasagar R, Sudarshan P, Ravindranath Singh N, Shivaram S. A rare presentation of an ectopic breast tissue in axilla. Our Dermatol Online. 2015;6(4):450-452.

Submission: 16.02.2015; **Acceptance:** 03.07.2015

DOI: 10.7241/ourd.20154.112

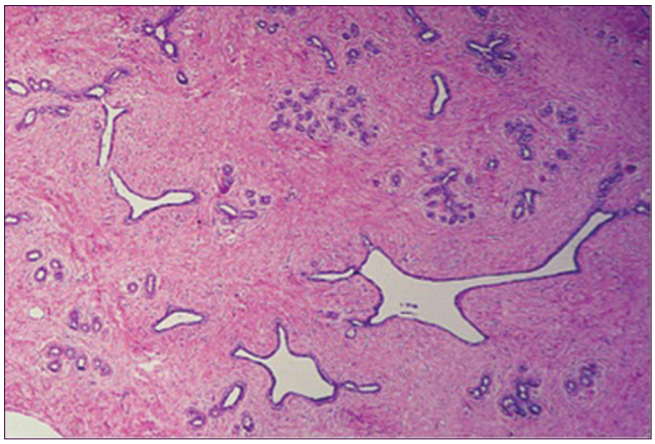


Figure 1: Microscopy picture of intracanalicular pattern of fibroadenoma.

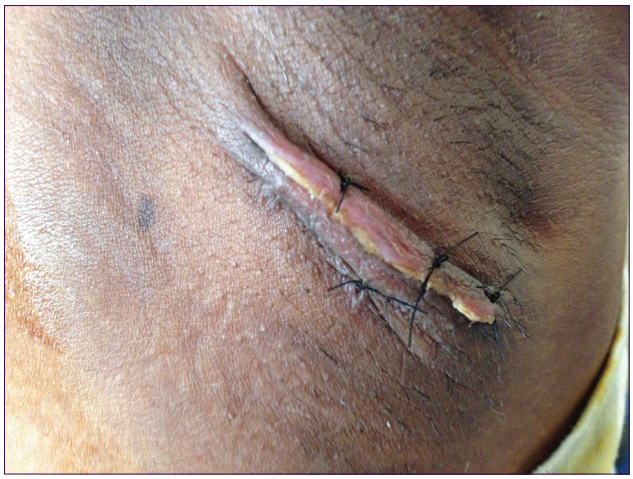


Figure 2: Post operative lession.

DISCUSSION

Polymastia is a term used to describe the presence of more than 2 breasts in human beings. It is synonymous with supernumerary or accessory breast tissue [1]. Accessory breast tissue has the potential to undergo the same benign and malignant changes as normal pectoral breast tissue.

The differential diagnosis for a solitary axillary mass may range from breast parenchymal lesions, lymph nodes, and infectious and vascular lesions, as well as an axillary tail of spence (extension of normal parenchyma towards the axilla) or a torn muscle belly. It is important to differentiate benign from malignant axillary masses to avoid unnecessary concern and intervention.

During the 6th week of embryonic development, the mammary milk lines, which represent 2 ectodermal thickenings, develop along the sides of the embryo, extending from the axillary region to the groin [1]. In normal development, most of the embryologic mammary ridges resolve, except for 2 segments in the pectoral region, which later become breasts.

Failure of any portion of the mammary ridge to involute can lead to ectopic breast tissue with (polythelia) or without (polymastia) a nipple-areolar complex. Therefore, ectopic breast usually occurs along the "milk line" or mammary line [2].

In 1915, Kajava published a classification system for supernumerary breast tissue that remains in use today. Class I consists of a complete breast with nipple, areola, and glandular tissue. Class II consists of nipple and glandular tissue but no areola. Class III consists of areola and glandular tissue but no nipple. Class IV consists of glandular tissue only. Class V consists of nipple and areola but no glandular tissue (pseudomamma). Class VI consists of a nipple only (polythelia) [1]. Class VII consists of an areola only (polythelia areolaris). Class VIII consists of a patch of hair only (polythelia pilosa). The accessory tissue may range from a subcutaneous focus of breast tissue to a full accessory breast complete with areola and nipple. The presence of a small nipple is the most frequently noted accessory breast structure.

The clinical significance of the polymastia lies in the fact that apart from the psychological and cosmetic impact, it develops the same pathological changes as the normally located breast tissue such as inflammation, fibrosis, fibroadenoma, cystosarcoma phyllodes, and carcinoma [3]. Diagnosis of EBT is strongly suggested by history of cyclic changes during the menstrual period or by initial appearance during pregnancy. Pathology in EBT should be evaluated by the same methods as in normal breast tissue. Radiological examination should be done to rule out the urogenital malformation as supernumerary kidneys, renal agenesis, and carcinomas [4].

Misdiagnosis of accessory breast tissue is common and the most common presumptive diagnoses include lipoma, lymphadenopathy, hidradenitis, sebaceous cyst, vascular malformation, and malignancy.

On mammography, accessory breast tissue in the axilla resembles normal glandular parenchyma and is separate from the breast, unlike the axillary tail of Spence, which is a normal direct extension toward the axilla from the main breast tissue.

The treatment of choice for symptomatic accessory axillary breast tissue is surgical excision. Cosmesis is the main indication in the majority of cases [5].

Removal of the tissue will relieve the physical discomfort and also confirms the diagnosis.

CONCLUSION

Fibroadenoma in the ectopic breast tissue is indeed a rare occurrence. It should also be considered as a differential diagnosis for all axillary swellings.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Amaranathan A, Balaguruswamy K, Bhat RV, Bora MK. An ectopic breast tissue presenting with fibroadenoma in axilla. Case Rep Surg. 2013;2013:947295.
2. Shin SJ, Sheikh FS, Allenby PA, Rosen PP. Invasive secretory (juvenile) carcinoma arising in ectopic breast tissue of the axilla. Arch Pathol Lab Med. 2001;1251372–4.
3. Rizvi G, Pandey H, Gupta MK. Fibroadenoma of ectopic breast tissue in axilla. J Case Reports. 2012;213-15.
4. Jethwani U, Saroha R, Verma R. Fibroadenoma of ectopic breast of axilla: A rare case report. Clin Canc Invest J. 2015;4:102-4.
5. Lesavoy MA, Gomez-Garcia A, Nejdl R, Yospur G, Syiau TJ, Chang P. Axillary breast tissue: clinical presentation and surgical treatment. Ann Plast Surg. 1995;35:356-60.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Parameatal urethral cyst of glans penis in children - A report of three cases

Mrinal Gupta[1], Anish Gupta[2]

[1]Sudhaa Skin Centre, Jammu, India, [2]Acharya Shri Chander College of Medical Sciences, Jammu, India

Corresponding author: Dr Mrinal Gupta, E-mail: drmrinalgupta@yahoo.com

ABSTRACT

Parameatal urethral cysts are a rare benign condition usually seen in males. They are usually asymptomatic but may produce symptoms like difficulty in micturition, pain during intercourse, urinary retention and distortion of the urinary stream. We report three cases of parameatal urethral cyst in young males presenting as a spherical clear fluid filled cystic lesions over the external urethral meatus, causing distortion of the urinary stream and poor cosmesis. Histological examination of the excised cyst showed a monolocular cyst lined with pseudo-stratified epithelium with no evidence of inflammation. Complete surgical excision of the cysts was done and no recurrence was observed at follow-up.

Keywords: Parameatal cyst; Glans penis cyst; External urethral meatus cyst

INTRODUCTION

Parameatal urethral cysts are a rare benign entity usually seen in males but rarely may be seen in females also. They were first described by Thompson and Lantin in 1956 and since then, around 50 cases have been reported in the literature [1,2]. These cysts are usually asymptomatic but may produce symptoms like difficulty in voiding, urinary retention, pain during intercourse or micturition, distorted urinary stream and poor cosmesis [2]. Herewith we report three cases of parameatal cyst in young males presenting with symptoms of distortion of urinary stream and poor cosmesis who were managed with complete cyst excision.

CASE REPORTS

Case 1

A 14 year old boy presented with a painless swelling over the glans penis which he noticed a year back. The swelling was asymptomatic in the beginning but for the last three months it was causing distortion of the urinary stream. There were no other symptoms other than distorted urinary stream and poor cosmesis. There was no history of trauma or application of topical medications. The boy gave history of needle aspiration of the lesion at some other centre six months back leading to its clearance at that time, but the lesion had recurred to its present size over the next six months. On examination, a smooth, spherical cystic mass about 0.8 cm in diameter was found at the external urethral meatus (Fig. 1). No inflammatory signs were present. On investigations, hematological counts, liver and renal function parameters were within normal limits. Urine microscopy and urine culture also revealed no abnormality. Complete excision of the cyst under local anesthesia was performed and good cosmetic results were obtained without any urinary flow problems. No recurrence was observed at six months of post-operative follow-up.

Case 2

A six year old male child presented to us with a three months history of a gradually progressive asymptomatic cystic swelling over the glans penis. There was no history of any preceding trauma or any surgical intervention. On examination, a well defined cystic

How to cite this article: Gupta M, Gupta A. Parameatal urethral cyst of glans penis in children – A report of three cases. Our Dermatol Online. 2015;6(4):453-455.
Submission: 21.03.2015; **Acceptance:** 12.05.2015
DOI: 10.7241/ourd.20153.123

swelling measuring 0.6cm in diameter was present over the glans penis without any inflammatory signs (Fig. 2). All the routine hematological and liver and renal function parameters were within normal range. Urine microscopy and urine culture also revealed no abnormality. From history and clinical examination a diagnosis of parameatal urethral cyst was made and complete excision of cyst under local anesthesia was done and no recurrence was observed over a post-operative follow-up period of eight months.

Case 3

A two year old child presented to us with an eight months history of development of an asymptomatic painless swelling over the glans. On examination, a well defined cystic swelling measuring 0.5 cm in diameter was present over the glans penis without any inflammatory signs. All the routine hematological and liver and renal function parameters were within normal range and urine microscopy and urine culture also revealed no abnormality. A diagnosis of parameatal urethral cyst was made a surgical deroofing of the cyst followed by phenolization of the cyst wall was done under general anesthesia and no recurrence was observed over a post-operative follow-up period of six months.

Histological examination of the excised cyst in both the cases showed a monolocular cyst lined with pseudo-stratified epithelium with no evidence of inflammation (Fig. 3).

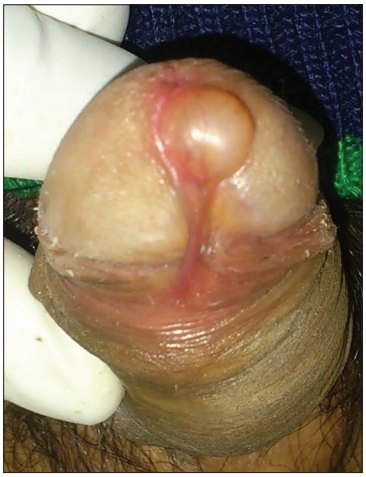


Figure 1: Parameatal cyst over the glans penis in a 15 year old male.

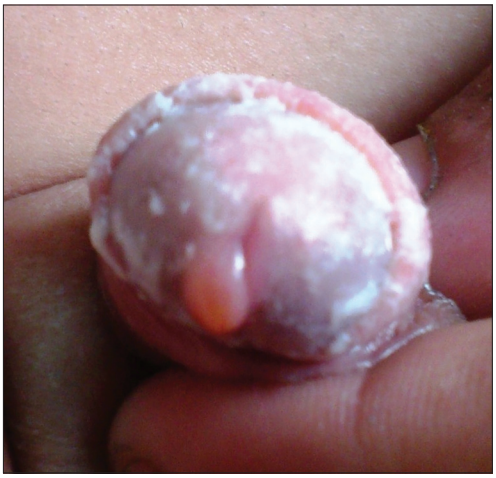


Figure 2: Parameatal cyst in a two year old child.

DISCUSSION

Parameatal urethral cysts are a rare entity usually seen in boys but cases among females have also been reported [3]. Usually they appear spontaneously during the second decade of life but congenital and infantile onset has also been seen [2]. The etiology of parameatal cysts is still not completely understood. Obstruction of paraurethral ducts, either spontaneous or secondary to infection, has been postulated as one possible factor by a few authors while others believe that parameatal cysts occur in the process of delamination or separation of the foreskin from the glands [2]. Ichiyanagi *et al* [4] detected the presence of Prostate specific antigen (PSA) in the cells of parameatal cysts, thus giving credibility to hypothesis that these cysts originate from the accessory male sex glands in the penile urethra. Soyer *et al* [3] reported two cases of parameatal cysts in newborn females, which were associated with vaginal bleeding and breast enlargement, which point towards the possible role of estrogens in their development.

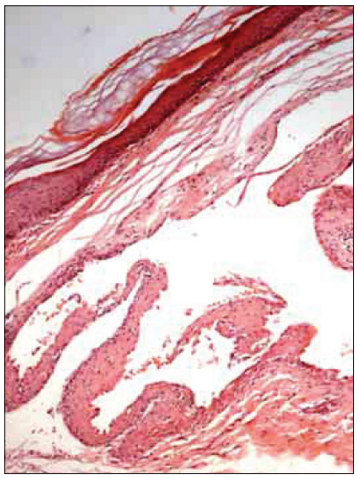


Figure 3: Histology of the excised cyst showing a monolocular cyst lined with pseudo-stratified epithelium without inflammation.

These cysts are usually small, measuring upto 1 cm in diameter. They usually occur on one side of the urinary meatus but bilateral cases have also been reported. Usually the parameatal cysts are asymptomatic but rarely may present with symptoms like difficulty in voiding, urinary retention, pain during intercourse or micturition, distorted urinary stream and poor cosmesis. If the cyst is traumatized, it may bleed, rupture or become infected [2].

Numerous treatment modalities like needle aspiration, marsupialisation, and decapping have been described in their management but complete surgical excision is the treatment of choice owing to lower chances of recurrence and better cosmetic results. Histologically, the cyst wall may be lined by columnar, squamous or transitional epithelium which varies according to the urethral segment of origin of the lesion [2,5].

In conclusion, a parameatal cyst is a benign, usually asymptomatic condition diagnosed on physical examination with a complete surgical excision providing good cosmetic results without recurrence.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Lantin PM, Thompson IM. Parameatal cysts of the glans penis. J Urol. 1956;76:753-5.
2. Lal S, Agarwal A. Parameatal Cyst: A Presentation of Rare Case and Review of Literature. J Clin Diag Res. 2013;7:1757-8.
3. Soyer T, Aydemir E, Atmaca E. Paraurethral cysts in female newborns: role of maternal estrogenes. J Ped Adol Gyn. 2007;20:249-51.
4. Ichiyanagi N, Shibata T, Matsumura T, Ishimoru H, Sakai I. Immunohistochemical identification of prostatic –specific antigen in parameatal urethral cysts of the glans penis. Br J Urol. 1998;81:170-1.
5. Aggarwal K, Gupta S, Jain VK, Goel A. Parameatal urethral cyst. Indian J Dermatol Venereol Leprol. 2008;74:430.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

The use of a topical compound cream product with chitosan, silver sulfadiazine bentonite hidrogel and lactic acid for the treatment of a patient with rosacea and ulcerated livedoid vasculopathy

Alin Laurentiu Tatu[1,2]

[1]*Faculty of Medicine and Pharmacy, University, Dunarea de Jos, Galati, Romania,* [2]*Private Practice, Galati, Romania*

Corresponding author: Dr. Alin Laurentiu Tatu, MD, PhD, E-mail: dralin_tatu@yahoo.com

ABSTRACT

Introduction: The aims of this study were to investigate the use of a topical compound cream product with Chitosan, Silver Sulfadiazine, Bentonite hidrogel and Lactic acid for the treatment of a patient with Rosacea and prolonged ulcerated Livedo Vasculitis. **Methods:** A patient with ulcerated Livedo Vasculitis applied the cream 22 months daily and was examined clinically. **Results:** At five months the area of the right foot ulcer decreased to a diameter of 18 mm. The pain disappeared (anamnestically) in 7 days around the ulcerated areas reticulated atrophic hipopigmented areas were seen-atrophie blanche and there ware still 4 small ulcerations. I noted also the disappearance of the livedoid area from the middle of the second toe finger. The left foot ulcer was epithelised.Wound cultures were negative. After another five months of continues use of the topical cream the situation improves, diameter of the ulcer decreased at 13 mm, the small ulcerations disappeared but continues to stay on the right foot. After another year the situation is stable at the right foot but the left ulcer was partially recurrent and it was painful. **Conclusions:** The topical compound formula with four active ingredients: Silver sulfadiazine, bentonite hidrogel, chitosan and lactic acid it was a cheap treatment for the patient, it was tolerated without sensitizing even it was used continuously for 22 months.

Key words: Livedoid vasculitis; Chronic ulcer; Topical compound cream; Silver sulfadiazine; Chitosan; Bentonite

INTRODUCTION

Livedoid vasculopathy is a disease of the lower legs with painful chronic ulcerations and cutaneous atrophy blanche located particularly around the malleoli and commonly seen on adult women. We present a case of a 39 years old man with Rosacea and dorsoplantar chronic and painful ulcerations known with the disease and referred to me for a topical treatment as he was partially refractory to systemic therapies and also to local ones. We used a compound topical cream product which contains chitosan, argentic sulfadiazine, bentonite and lactic acid with good result and tolerance. The ulceration decreased and the pain was disappeared.

CASE REPORT

A 39 years old man was referred to my Private Practice from the Vascular Surgery unit with a long history -7 years- of painful ulcerations on the dorsoplantar areas and sometime around maleoli. The patient was previously consulted those years by dermatologists, vascularsurgeons, internists and the Diagnosis of livedoid vasculopathy was established before but now he said is the most painful period and he needed help for the wound to heal. The patients history reveals Rosacea and hypercholesterolemia. Previously he was treated in time with low –dose aspirin, dipyridamole, pentoxyphylline, fraxiparine. At the time of the referral he was on treatment with rosuvastatin 20mg daily and clopidogrel 75 mg daily, he used topically

How to cite this article: Tatu AL. The use of a topical compound cream product with chitosan, silver sulfadiazine bentonite hidrogel and lactic acid for the treatment of a patient with rosacea and ulcerated livedoid vasculopathy. Our Dermatol Online. 2015;6(4):456-459.

Submission: 01.06.2015; **Acceptance:** 28.07.2015

DOI: 10.7241/ourd.20154.124

on ulcers silver dressings and for the face metronidasole cream. He had no history of smoking or of thrombosis and no varicous visible veins. Physical examination revealed one large (66mm of diameter) painful ulceration on dorsoplantar area of the right foot, with some edema and circular erythema around it and also a livedoid area on the second toe finger (Fig. 1). On the surface of the ulcer there was a detritus with the rest of previous treatments,pus and scales. The second ulceration was in perimaleolar area of the foot (8 mm of diameter) (Fig. 2). Wound cultures from both ulcers showed *Klebsiella* with maximum sensitivity to Cefixime. Venous ultrasound was normal. He had two previous biopsys consistent with stasis, fibrosis, hyalinization of the vessels, the PAS (Periodic Acid Schiff) taining was negative, thrombosis of the superficial vessels, red blood cell extravasation suggesting the diagnosis of livedoid vasculopathy. The tests for Protein C, Protein S, homocysteine, antithrombine III, Lupusanticoagulant, Cryoglobulines, Cholesterol, Anticardiolipine, antibodies, anti –double-stranded DNA antibodies, complement, prothrombin time/ partial thromboplastin time, aspartate aminotransferase/ alanine aminotransferase,were within normal limits or negative [1]. The Erythrocyte sedimentation rate was elevated at 42 mm/hr.

We added at his previous treatment Cefixime 400 mg daily for 10 days and after the informed consent was signed and the Institutional Review board approval was taken, we advise the patient to apply once a day a combination product containing chitosan, argenticsulfadiazine, bentonite and lactic acid (Cicatrol, AntibioticeIasi, Romania). At five months the area of the right foot ulcer decreased and there were still 4 ulcers but just one with a diameter of 16mm.The pain disappeared (anamnestically) in 7 days –so it was maybe due to infection. Around the ulcerated areas reticulated atrophic hipopigmented areas were seen-atrophied blanche (Fig. 3). We noted also the disappearance of the livedoid area from the middle of the second toe finger. The perimaleolar foot ulcer was epithelized (Fig. 4). Wound cultures were negative. After another five months of continuous use of the topical cream the situation improves, the other three small ulcerations disappeared, the big one diminished at 12 mm but continues to stay on the right foot (Fig. 5).

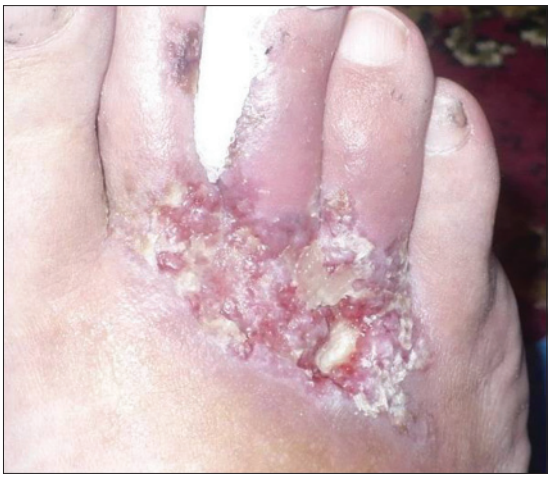


Figure 1: Painful ulceration on dorsoplantar area of the right foot.

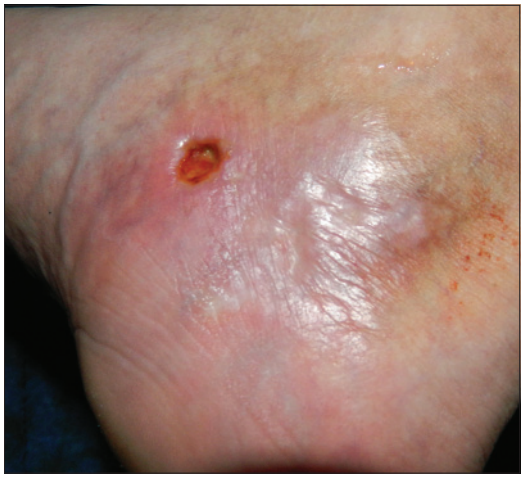


Figure 2: Ulceration of perimaleolar area of the foot.

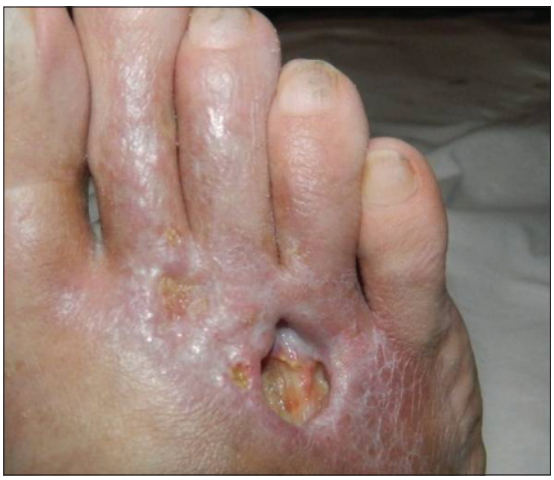


Figure 3: Decreased ulceration of dorsoplantar area of the right foot after 5 months.

DISCUSSIONS

Livedoid vasculopathy is a disease of the lower legs with painful chronic ulcerations and cutaneous atrophy blanche, it is hard to manage due to the resistance at topical and internal treatments and its potential to complicate with infections.Topical creams has

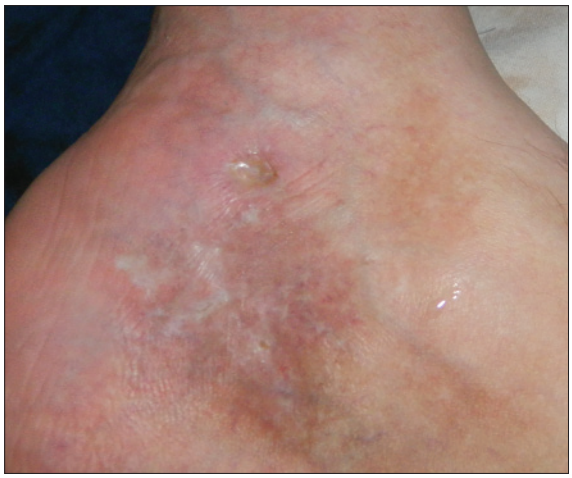


Figure 4: Perimaleolar ulcer is epithelised after 5 months.

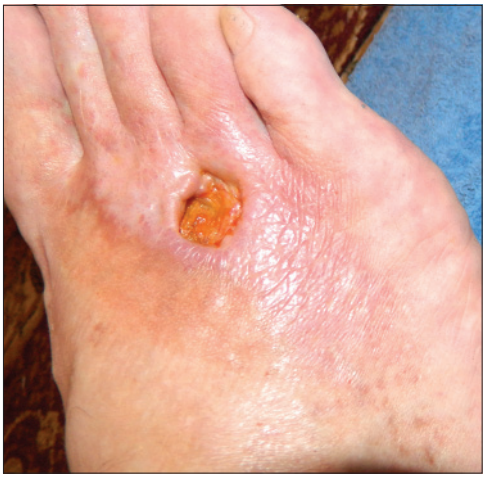


Figure 5: The dorsoplantar ulceration after 10 months.

sensitizing potential when used for long periods of time. The topical cream formula contains four active ingredients: Silversulfadiazine, bentonitehidrogel, chitosan and lactic acid.

Silver is used to reduce infections from ancient times. The use of silver impregnated dressings after laminectomies appear to limit/reduce the incidence of both postoperative deep and superficial wound infections [2].The wide spectrum of antibacterial activity the low toxicity, minimal tissue reaction, ease of application suggest that topical silver sulfadiazine can safely be used in burns, surgical wounds and can be extended to other wound infections, wound covers and some transplant materials [3]. At the contact with the bacteria cell Ag + penetrates the bacterial membrane and stop the DNA synthesis so the bacteria cannot divide so it dies. Due to his redox potential Ag+ dissociate after the bacterialdestruction and acts in a bactericide manner ensuring a cyclic process. Silver sulfadiazine have excellent anti-bacterial activity and could be used initially while the identification of the infective agent is required for selecting the alternative topical agents and/or systemic therapy [4].The second ingredient, bentonite is a clay which by absorbing or adsorbing moisture and impurities from the skin serve to cleanse and to refresh the surface of the skin surface and to aid the healing process. External use of Bentonite for wound healing is safe and feasible and the macroscopic healing of the wounds treated by Bentonite was superior versus control group [5]. The third ingredient is Chitosan- a polysaccharide obtained from crustaceans and with a regenerative effect on skin.Chitosan is a linear copolymer of B linked 2-acetamido-2 deoxy-B-D-glucopyranose and 2-amino-2-deoxy-B-Dglycopyranose,easily obtained by deacetilation of chitin, an abundant polysaccharide found in nature as a component of exoskeletons of crustaceans and insects. Topically used Chitosan bind on fibroblasts and stimulates the keratinocite proliferation and the repair of the epiderm [6]. Since its discovery approximately 220 years ago, Chitosan as a cationic natural polymer has been used as topical dressing in wound management owing to its hemostatic, stimulation of healing, antimicrobial, nontoxic, biocompatible and biodegradable properties [7]. It stimulates the imunocites and also leucocites, macrophages and fibroblasts [8], bloks nerve endings to reduce pain, absorbe fluids from inflammation, encourages natural blood clotting, forms barrier against infection, provides proteins for healing and scaffold for cell growth, strengthens new tissue, minimize scarring [9].

The protection of the host against bacterial infection is stimulated by chitosan. The effectiveness of chitosan bacteriastatic properties were tested against bacterial strains and a common skin fungus. Powered chitin, chitosan or whole crab shells were not effective in any of the tests, but solution of chitosan in acetic acid inhibited the bacterial strains and the fungus. The mechanism underlying the inhibition of bacterial growth, is though to be that the cationically charged amino-group may combine with anionic components such as N-acetylmuramic acid, sialic acid and neuraminic acid, on the cell surface, and may suppress bacterial growth by impairing the exchanges with the medium, chelating transition meal ions and inhibiting enzymes. Chitosan is characterized by high antibacterial and fungicidal activities [10]. The lactic acid is known as a antiwrinkle agent,hepls

to the renewing of epiderma, stimulates the colagen sinthesis,ceramides and epidermal growing factor [11].

The associated Rosacea was not influenced by the evolution of the ulcers and i didn't find in literature the association with livedoid vasculopathy.

CONCLUSIONS

Livedoid vasculopathy is a disease of the lower legs with painful chronic ulcerations and cutaneous atrophy blanche. livedoid vasculopathy is a coagulation disorder. As a result, modern therapy strategies take into account to treat it with systemic anticoagulation. Topical therapies may be considered only as accompanying measures but very important.

It is hard to manage due to the resistance at topical and internal treatments-fibrinolitics, anticoagulants, anti-plateletagregants- and due it is potential to complicate with infections.Topical creams has sensitizing potential when used for long periods of time. The topical compound formula with four active ingredients: Silver sulfadiazine, bentonitehidrogel, chitosan and lactic acid it was a cheap treatment for the patient, it was tolerated without sensitizing even it was used continuously for 10 months. Unfortunately one ulcer didn't close completely. Our experience was limited to one case of livedoid vasculopathy and of course more studies should be done in the future on this field.

AKNOWLEDGEMENTS

This paper is supported by the Sectoral Operational Programme Human Resources Development (SOP HRD), financed from the European Social Fund and by the Romanian Government under the contract POSDRU/159/1.5/S/137390.

Consent

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Trey Haunson G, David J, Prall N, Miller R. Livedoid Vasculopathy: Review of Pathogenesis, Clinical Presentation, Diagnostic Workup, and Treatment. Cutis. 2012;90:302-6.
2. Epstein NE. Do silver impregnated dressings materials limits infections after lumbar laminectomy with instrumented fusion?. Surg Neurol. 2007;68:483-5,discussion 485.
3. Fox CL Jr. Topical therapy and the development of silver sulfadiazine. Surg Ginecol Obstet. 1983;157:82-8.
4. Katara G, Chamania S, Chitnis S, Hemvani N, Chitnis V, Dahananjai SC. A comparative study of the effect of different topical agents on burn wound infections. Indian J Plast Surg. 2012;45:374-78.
5. Emami Razavi SH, Esmaeili N, Forouzania SK, Amanpour S, Rabbani S, Alizadeh AM, et al. Effect of Bentonite on skin wound healing: experimental study in the rat model. Acta Medica Iranica. 2006;44:235-40.
6. Chatelet C, Damour O, Domard A. Influence of the degree of acetylation on some biological properties of chitosan films. Biomaterials. 2001;22:261-8.
7. Dai T, Tanaka M, Huang YY, Hamblin MR. Chitosan preparations for wound and burns: antimicrobial and wound healing effects. Expert Rev Anti Infect Ther. 2011:9:857-9.
8. Aranaz I, Mengibar M, Harris R, Panos I, Miralles B, Acosta N, Galed G, Heras A. Functional Caracterisation of Chitin and Chitosan. Current Chem Biol. 2009;3:203-30.
9. Paul Wan, Sharma CP. Chitosan and Alginate Wound Dressings: A Short Review. Trends Biomater Artif Organs. 2004;18:18-23.
10. Balicka-Ramisz1 A, Wojtasz-Pajak A, Pilarczyk B, Ramisz1 A, Laurans1 L. Antibacterial anf antifungal activity of Chitosan. ISAH 2005 - Warsaw, Poland, Vol 2.
11. Kim SJ, Park JH, Kim DH, Won YH, Maibach HI. Increased in vivo collagen synthesis and in vitro cell proliferative effect of glycolic acid. Dermatol Surg. 1998;24:1054-8.

Source of Support: European Social Fund and by the Romanian Government under the contract POSDRU/159/1.5/S/137390. **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Isotretinoin induced rash, urticaria, and angioedema: A case report

Zonunsanga

Department of Skin and VD, RNT Medical College, Udaipur, Rajasthan-313001, India

Corresponding author: Dr. Zonunsanga, E-mail: jrkos04@gmail.com

ABSTRACT

Isotretinoin is a vitamin A analogue, which is readily isomerized to tretinoin. It causes normalization of abnormal keratinisation. It also reduces sebum secretion. It also has anti-inflammatory as well as antibacterial properties. It has some adverse effects like teratogenecity, hypertriglyceridemia, pancreatitis, dryness of skin, cheilitis, altered liver functions etc. A 25 years old unmarried lady presented with acne vulgaris, who did not showed improvements with conventional (antibiotics) therapy was given isotretinoin. She developed maculopapular rash, urticaria and angioedema Isotretinoin induced urticarial rashes and angioedema is rarely reported as far as our knowledge is concerned.

Key words: Isotretinoin; acne vulgaris; urticaria; angioedema

INTRODUCTION

Isotretinoin is a vitamin A analogue, which is readily isomerized to tretinoin. It causes normalization of abnormal keratinisation [1-4]. It also reduces sebum secretion.It also has anti-inflammatory as well as antibacterial properties [1-5]. It has some adverse effects like teratogenecity, hypertriglyceridemia, pancreatitis, dryness of skin, cheilitis,altered liver functions etc. Isotretinoin induced urticarial rashes and angioedema is rarely reported as far as our knowledge is concerned [6-9].

CASE REPORT

A 25 years old unmarried lady presented with acne vulgaris for which she was prescribed oral Doxycycline 100 mg bd along with topical clindamycin gel and topical benzoyl peroxide 2.5% gel. Since there was no significant response even after 2 months, doxycycline was stopped and azithromycin pulse therapy (Azithromycin 500 mg od for 3 days, repeat the same after every 10 days) for 1 month along with the topical medications which were given previously, which did not show much improvement. Finally she was given Isotretinoin (after liver fuction test, Haemogram and lipid profiles were done and proved to be within normal range). After 5 days of medication, she started developing discrete to confluent, maculopapular rashes over bilateral upper extremities (Fig. 1), which was progressed to involve face (Fig. 2), neck (Figs 3 and 4), trunk and bilateral lower extremities (Fig. 5) within a period of 2-3 days. It was also associated with urticaria in some places, which was persisted for more than 24 hrs, along with oedema of face including lips, suggestive of angioedema. She was advised to stop all medications and was given oral prednisolone 2 mg for 7 days along with oral antihistamines. The urticaria and rashes began to resolve 3 days of starting prednisolone and complete clearance of the lesions took around 2-3 weeks. Provocation was not done as the patient reacted with urticaria and angioedema.

DISCUSSION

Isotretinoin is a first generation oral retinoid.It binds to the retinoid receptors. The retinoid receptors are of two families, RAR and RXR. RAR are paired with RXR whereas RXR may form homodimer with one another, or heterodimer with other receptors [1]. Upon binding of ligand, the receptor complex acts

How to cite this article: Zonunsanga. Isotretinoin induced rash, urticaria, and angioedema: A case report. Our Dermatol Online. 2015;6(4):460-462.
Submission: 06.01.2015; **Acceptance:** 09.05.2015
DOI: 10.7241/ourd.20153.125

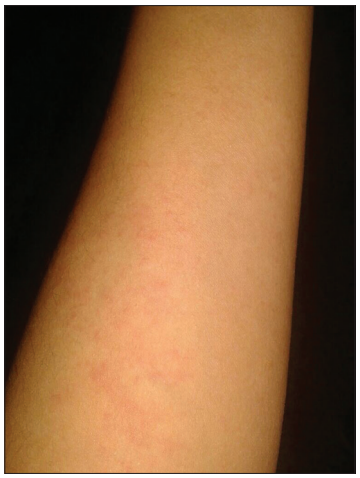


Figure 1: Maculopapular rash over dorsum of hands.

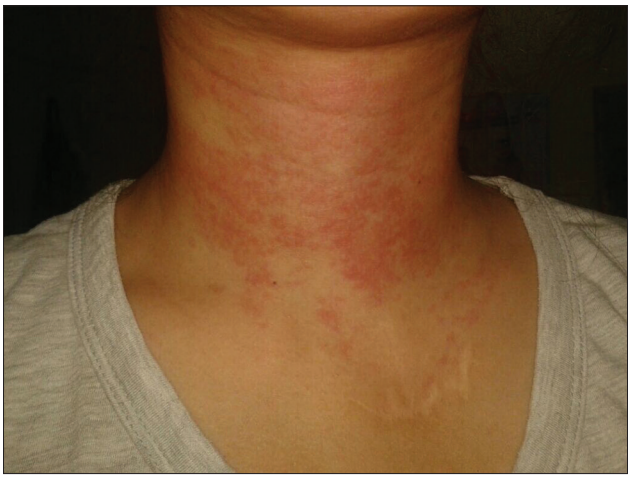


Figure 2: Maculopapular rash over face

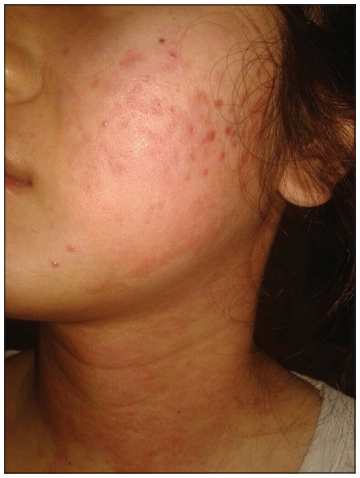


Figure 3: Maculopapular rash over neck.

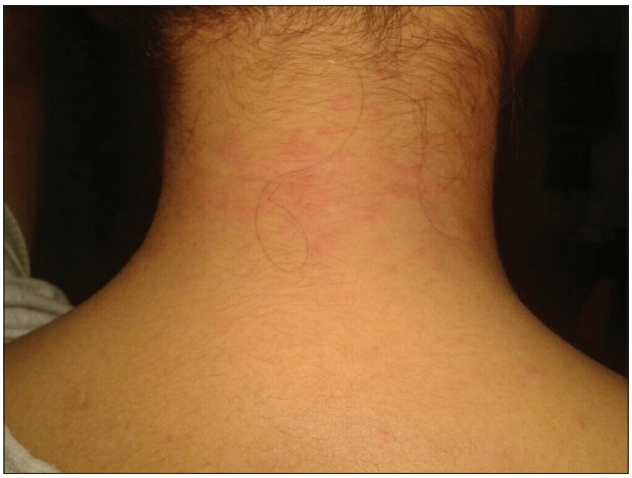


Figure 4: Maculopapular rash over neck.

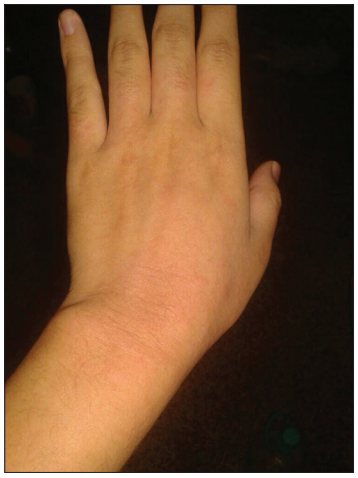


Figure 5: Maculopapular rash over legs.

as transcription factor as well as antagonist of other transcription factors [1,6,7,10]. The resultant actions includes normalization of keratinization by regulating cell growth, differentiation and morphogenesis of keratinocytes; decreases cell cohesiveness causing comedolytic and antikeratinizing effects; immunomodulatory and antiinflammatory actions; decreasing sebum production; antibacterial action by reducing follicular space and nutritional supply of Propionibacterium acne [4,7,9,12]. The oral bioavailability may be enhanced with food.It has half life of 10-20 hours, and completely cleared from the body with 1 month of stoppage of the drug. Some common adverse effects include teratogenecity, reduced night vision, dry eyes, lipid profile abnormalities especially hypertriglyceridemia, pancreatitis, altered liver functions, agranulocytosis and raised intracranial pressure [2,12]. Sweet's syndrome, hepatosplenomegaly, lymphadenopathy, myalgia, vasculitis, arthritis and inflammatory bowel diseases were also reported [6-8]. Besides, vitamin B 12 and folic acid level in the serum might be decreased after isotretinoin therapy [9]. There were some reports of association with myocardial

infarction, stroke and thromboembolic events with isotretinoin [10]. In addition, nail changes like transverse leuconychia had been reported [11]. The FDA approved indications include nodulocystic acne and recalcitrant acne. Other off-label indications include follicular disorders like rosacea, Hirradenitis suppurativa etc; disorders of keratinization; Viral infection like Human papilloma virus infection [12]. It has category X status for pregnancy.

REFERENCES

1. Rao PK, Bhat RM, Nandakishore B, Dandakeri S, Martis J, Kamath GH. Safety and efficacy of low-dose isotretinoin in the treatment of moderate to severe acne vulgaris. Indian J Dermatol. 2014;59:316.
2. Kızılyel O, Metin MS, Elmas OF, Cayır Y, Aktaº A. Effects of oral isotretinoin on lipids and liver enzymes in acne patients. Cutis. 2014;94:234-8.
3. Cunha Filho RR, Almeida Jr HL, Breunig Jde A. Angiodema due to oral acitretin and isotretinoin. An Bras Dermatol. 2011;86(4 Suppl 1):S28-30.
4. Saray Y, Seçkin D. Angioedema and urticaria due to isotretinoin therapy. J Eur Acad Dermatol Venereol. 2006;20:118-20.
5. Barzilai A, David M, Trau H, Hodak E. Seborrheic dermatitis-like eruption in patients taking isotretinoin therapy for acne: retrospective study of five patients. Am J Clin Dermatol. 2008;9:255-61.
6. Moghimi J, Pahlevan D, Azizzadeh M, Hamidi H, Pourazizi M. Isotretinoin-associated Sweet's syndrome: a case report. Daru. 2014;22:69.
7. Leibovitch I, Amital H, Levy Y, Langevitz P, Shoenfeld Y. Isotretinoin-induced adult onset Still's disease. Clin Exp Rheumatol. 2000;18:616-8.
8. Rashtak S, Khaleghi S, Pittelkow MR, Larson JJ, Lahr BD, Murray JA. Isotretinoin Exposure and Risk of Inflammatory Bowel Disease. JAMA Dermatol. 2014;150:1322-6.
9. Gökalp H, Bulur I, Gürer M. Decreased vitamin B12 and folic Acid concentrations in acne patients after isotretinoin therapy: a controlled study. Indian J Dermatol. 2014;59:630.
10. Lorenzo N, Antuña P, Dominguez L, Rivero F, Bastante T, Alfonso F. Acute myocardial infarction in a young woman on isotretinoin treatment. Int J Cardiol. 2014;181C:39-41
11. Gregoriou S, Banaka F, Rigopoulos D. Isotretinoin-induced transverse leuconychia. J Eur Acad Dermatol Venereol. 2014 Oct 29.
12. Nickle SB, Peterson N, Peterson M. Updated Physician's Guide to the Off-label Uses of Oral Isotretinoin. J Clin Aesthet Dermatol. 2014;7:22-34.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Case Report

Skin reaction to bed bugs bite reflecting erythema multiforme: Case report

Michał Andres[1], Andrzej Jaworek[1], Tomasz Stramek[2], Anna Wojas-Pelc[1]

[1]Department of Dermatology, Jagiellonian University College of Medicine, Krakow, Poland, [2]Student Scientific Association of the Department of Dermatology, Jagiellonian University College of Medicine, Krakow, Poland

Corresponding author: Dr. Michał Andres, E-mail: mandres@o2.pl

ABSTRACT

Bed bugs (Cimex spp.) are wingless, hematophagous arthropods causing bites, which generates wide range of skin reaction and may be misdiagnosed with more serious disease as dermatitis herpetiformis, bullous pemphigoid or erythema multiforme. Differential diagnosis could be a challenge especially in western countries where bed bugs were forgotten since the Second World War. There are only a few reports of serious anaphylaxis after bed bugs exposition and the evidences about the role of bed bugs as vectors to some infectious diseases are not conclusive, but bites could be a source of physical ailments and psychological distress. We present a case of 24 year old female patient who had been bitten by bed bugs and primarily diagnosed with erythema multiforme. After releasing from hospitalization because of successful treatment patient developed another eruption of skin lesions, this time more characteristic to bed bugs bites during one day. The new diagnosis of bed bugs bites was confirmed with proving the presence of these arthropods in her rented apartment.

Key words: Bed bug bites; Bed bug reactions; Erythema multiforme

INTRODUCTION

Bed bugs (Cimex spp.) are wingless, oval, flattened insects that grow up to 5-6 mm in length. The adults are deep brown in color while immature are smaller and yellow to brown (Fig. 1). Since the mid 1990s we notice a global resurgence of these hematophagous arthropods. Patients suffering from bed bugs bites become more frequent every year and may be misdiagnosed, because clinical reactions to bites are not specific and may resemble many other dermatological diseases such as erythema multiforme, urticaria or even bullous pemphigoid [1,2]. Bed bugs are not known to transmit infectious diseases but their interactions with hosts could be a source of physical ailments and psychological distress. Polish medical staff should be aware of this misleading source of skin lesions since the problem of infestations might grow over time.

CASE REPORT

A 24 year old women was admitted to the Department of Dermatology of the University Hospital in Krakow because of suspicion of erythema multiforme. Three weeks before patient had presented skin reaction consisting of vesicles and bullae on erythematous-edematous base. After another two days an edema of the right ankle had been noticed. Local general practitioner put patient on glucocorticoids (methylprednisolone in 4-12 mg daily dosage). She neglected any chronic diseases, allergies, operations and chronic medication despite of birth control pills (0,02 mg ethinyl estradiol + 3 mg drospirenone). Her family history was unspecific. Patient was admitted in good general state and presented itching, erythematous-edematous lesions with vesicles on top of some lesions on skin of her legs and arms. Some of lesions were scratched.

How to cite this article: Andres M, Jaworek A, Stramek T, Wojas-Pelc A. Skin reaction to bed bugs bite reflecting erythema multiforme. Case report. Our Dermatol Online. 2015;6(4):463-465.

Submission: 08.08.2015; **Acceptance:** 12.09.2015

DOI:10.7241/ourd.20154.126

Figure 1: *Cimex lectularius* found by patient's partner in rented apartment

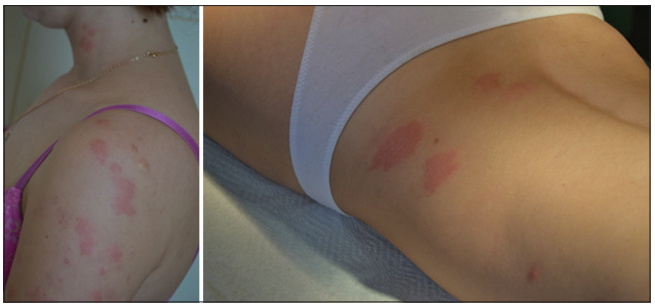


Figure 2: A 24 year old patient with multiple, erythematous-edematous lesions localized on the skin of the face, neck, lower back, arms and buttocks after bed bugs bites

The laboratory tests and urinalysis did not reveal any important abnormalities. Her glucose, urea, creatinine, electrolytes, ALT, ASP, bilirubin, complete IgE levels in blood were normal. There was slightly decreased level of C1 inhibitor in blood but the level of C3c and C4 ranged within regular laboratory limits. Histopathology of the dermis and epidermis specimen was described as orthokeratotic, spongiotic and of normal granulosis. There were lymphocyte and eosinocyte infiltration around the superficial vascular plexus and signs of fibrosis in the dermis.

The patient was treated with methylprednisolone (12 mg per day), rupatadine (10 mg daily) and clobetasone ointment. After three days of hospitalization was released from the hospital because of complete remission of skin lesions. Next day patient came back to the Department of Dermatology and presented an eruption of new, multiple, erythematous-edematous lesions localized on the skin of the face, neck, lower back, arms and buttocks (Fig. 2). Linear formation of some lesions could be observed. Patient's partner presented similar lesions. After taking an expanded patient history she was admitted to the hospital again and the diagnosis of severe skin reaction to bed bugs bite were made. Patient's partner was instructed to do thorough search of rented apartment to confirm the diagnosis. After all-night search he brought two adult specimens of bed bugs and the causative agent of the disease was confirmed.

DISCUSSION

In temperate climate the most common bed bugs are *Cimex lectularius*, All *Cimex* species have mouthparts adjusted to pierce and suck mammal or bird blood. They penetrate the skin and inject anticoagulants and other pharmacologically active substances like kinins or proteases and then suck blood with other liquefied tissues. Cutaneous reaction to bed bugs bites include erythema, wheals or vesicle formation[1,2]. Patient's immunocompetence and previous exposure to the bites affects the type of reaction. Systematic exposure can sometime lead to purpuric macules as an only sign of the bite[3]. There are some papers on systemic reactions from bed bug bites[4], but a recent study by Reinhardt revealed that up to 45% of exposed people presented no skin reaction on the first bite. Cleary, C. and Buchanan, D. categorized dermatological reaction to bed bugs bites into four types: 1. papular lesions grouped in a linear formations ("breakfast, lunch, and dinner" sign), 2. pruritic wheals, 3. small vesicles on erythematous base, 4. bullous lesions of the hands and feet similar to erythema multiforme[5]. Lesions tends to localize on the uncovered parts of the body like face, neck, arms or legs. The clinical consequences of bed bugs infestations include mental health issues like delusions or sleeplessness [1]. Scratching can lead to secondary skin infections[6]. There are some evidence that bed bugs could be a vector to some infectious diseases as HBV[7], but no evidence of transmission of HCV or HIV has been made[8,9]. Histology is unspecific as in any other arthropod bites and may include lymphocyte and eosinocyte perivascular infiltration[10].

Taking a detailed patient history involving home environment, work conditions and contact with domestic animals could be very important to diagnose reaction to bed bug bites. A thorough inspection of home environment in useful [11]. Differential diagnosis could be a challenge especially in western countries where bed bugs were forgotten since the Second World War because of spread of use of pesticides,

but nowadays, when pesticides are less toxic and milder bed bugs infestations may occur more often. Cohen et al. included in the differential diagnosis diseases like erythema multiforme, atopic dermatitis, scabies, lice infestations, papular urticaria, dermatitis herpetiformis, bullous pemphigoid, angioedema and other arthropods bites[12].

Untreated lesions tends to disappear after 1 to 2 weeks [7]. Symptomatic treatment involving topical steroids and systemic antihistamines should be implemented. Some more serious cases may require the use of systemic steroids [13]. Secondary infections may lead to topical or systemic antibiotic therapy[14]. There are multiple review papers covering the deterrence and prevention of bed bug infestations[2,7,13], but in authors opinion the qualified extermination company should be engaged if the problem affects home environment of patient. There are some preventive measures that can protect people against exposition while travelling including checking hotel rooms for signs of infestation, moving the bed away from the wall and keeping bed linen away from the floor or checking the luggage for signs of bed bugs presence before coming home.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1 Elston DM, Stockwell S. What's eating you? Bedbugs. Cutis. 2000;65:262–4.
2 Harves AD, Millikan LE. Current concepts of therapy and pathophysiology in arthropod bites and stings. Part 2: Insects. Int J Dermatol. 1975;14:621–34.
3 Sansom JE, Reynolds NJ, Peachey RD. Delayed reaction to bed bug bites. Arch Dermatol. 1992;128:272–3.
4 Bircher AJ. Systemic immediate allergic reactions to arthropod stings and bites. Dermatology. 2005;210:119-27.
5 Cleary CJ, Buchanan D. Diagnosis and management of bedbugs. Nurse Practit. 2004;29:47–8.
6 Burnett JW, Calton GJ, Morgan RJ. Bedbugs. Cutis. 1986;38:20.
7 Silverman AL, Qu LH, Blow J, Gordon SC, Walker ED. Assessment of hepatitis B virus DNA and hepatitis C virus RNA in the common bedbug (Cimex lectularis L. and kissing bug (Rodnius prolixus). Am J Gastroenterol. 2001;96:2194-8.
8 Mayans MV, Hall AJ, Inskip HM. Do bedbugs transmit hepatitis B? Lancet. 1994;343:761-3.
9 Adler MW. Development of the epidemic. BMJ. 2001;322:1226-9.
10 Parsons DJ. Bedbug bite anaphylaxis misinterpreted as coronary occlusion. Ohio State Med J. 1955;51:669.
11 Thomas I, Kihiczak GG, Schwartz RA. Bedbug bites: A review. Int J Dermatol. 2004;43:430–3.
12 Cohen PR, Tschen JA, Robinson FW, Gray JM. Recurrent episodes of painful and pruritic red skin lesions. Am J Clin Dermatol. 2010;11:73-8.
13 Goddard J, deShazo R. Bed bugs (Cimex lectularius) and clinical consequences of their bites. J Am Med Assoc. 2009;301:1358–66.
14 Honig PJ. Arthropod bites, stings, and infestations: their prevention and treatment. Pediatr Dermatol. 1986; 3:189–97.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Review Article

Hepatitis C in dermatology

Zonunsanga

Department of Skin and VD, RNT Medical College, Udaipur, Rajasthan-313001, India

Corresponding author: Dr. Zonunsanga, E-mail: jrkos04@gmail.com

ABSTRACT

Hepatitis C is a serious public health problem all over the world. It is caused by a single stranded RNA virus. Most acute infections are subclinical, but in 75% of individuals, infection leads to a chronic hepatitis, which in some cases can progress to cirrhosis and occasionally development of hepatoma. It has wide range of dermatological manifestations. This review article deals with the overview of epidemiology, pathogenesis, clinical manifestations, management and prevention.

Key words: Hepatitis; Epiphenomenon; Interleukins; Interferon

INTRODUCTION

Hepatitis C is a serious public health problem all over the world. So far, it has been reported to infect around 170 million people worldwide. Hepatitis C virus is a linear, single-strand, positive-sense, 9600-nucleotide RNA virus, the genome of which is similar in organization to that of flaviviruses and pestiviruses; HCV is the only member of the genus *Hepacivirus* in the family Flaviviridae. The virus replicates in hepatocytes and blood mononuclear cells. Most acute infections are subclinical, but in 75% of individuals, infection leads to a chronic hepatitis, which in some cases can progress to cirrhosis and occasionally development of hepatoma. A variety of conditions ranging from endocrinopathies to different skin diseases have been described in HCV infections [1-7].

EPIDEMIOLOGY

Prevalence

Around 170-200 million infected individuals worldwide, 3-5 million in the USA. An estimated 10-30 of all those who are infected may develop major liver damage in the 15-30 years following contracting the infection. The prevalence of hepatitis C is lowest in Northern European countries, including Great Britain, Germany and France in which the prevalence of HCV antibodies in blood donors averages less than 1% for the regions whereas in the U.S, it is approximately 2.5%. Higher rates have been reported in Southeast Asian countries, including India (1.5%), Malaysia (2.3%), and the Philippines (2.3%) [1-7].

Race

Race and ethnicity do not relate to hepatitis C virus (HCV). HCV infection is associated with lower economic status, less education and groups other than whites [1-7].

Sex

No sex preponderance occurs with hepatitis C virus (HCV) infection. Sex differences were not significant [1-7].

Age

About 65% of individuals positive for hepatitis C virus (HCV) antibodies are aged 30-49 years. Younger age at infection often relates to lesser consequences of the infection [1-7].

Transmission

The virus is being transmitted by [1-8]:

1. Blood transfusion,

How to cite this article: Zonunsanga. Hepatitis C in dermatology. Our Dermatol Online. 2015;6(4):466-470.
Submission: 06.11.2014; **Acceptance:** 06.04.2015
DOI: 10.7241/ourd.20154.127

2. Percutaneous routes, such as injection drug use.
3. Occupational exposure to blood and the likelihood of infection is increased in hemodialysis units.
4. It can be transmitted sexually
5. Perinatally

About 10–15% of patients with acute hepatitis C report having potential sexual sources of infection. The chances of sexual and perinatal transmission have been estimated to be 5%. Breast-feeding does not seems to increase the risk of HCV infection between an infected mother and her infant. Health workers are more likely to acquire HCV infection through accidental needle punctures, the efficiency of which is 3%. Infection of household contacts is rare as well [1-8].

High-risk groups [2,3,5-9]:

1. Persons who have used injection drugs or those who have used illicit drugs by non injection routes
2. Persons with HIV infection
3. Hemophiliacs treated with clotting factor concentrates
4. Hemodialysis patients
5. Persons with unexplained elevations of aminotransferase levels
6. Transfusion or transplantation recipients
7. Children born to women with hepatitis C
8. Health care, public safety and emergency medical personnel following needle injury or mucosal exposure to HCV-contaminated blood
9. Sexual partners of persons with hepatitis C infection.

PATHOGENESIS

Different factors such as viral, genetic or environmental may be responsible for cutaneous disorders associated with HCV infection. In most cases, the mechanisms through which HCV may trigger or exacerbate skin manifestations remain unclear and require further examinations.

The pathomechanism of various dermatologic manifestations of HCV can be classified into the following main types [1,8-11]:

1) Primary due to direct HCV infection of the skin, lymphocytes, dendritic cells and blood vessels. This hypothesis has been confirmed by the detection of HCV RNA particles in epidermal cells and skin lesions.
2) Skin manifestations of HCV infection may be an epiphenomenon resulting from the interruption of immune responses by interfering with host T-cell function due to down-regulating interleukin 2 and interferon gamma function and up-regulating interleukin-10. An example would be cryoglobulinemia-induced leukocytoclastic vasculitis.
3) Disruption of HCV-infected organs other than skin may produce nonspecific cutaneous signs due to typical skin responses to that organ. For example, thyroid hormone release in early HCV-linked autoimmune thyroiditis can culminate in skin responses and manifestations.
4) Neoplastic dermatologic manifestations are another category of extrahepatic findings. Local carcinogenic functions of HCV, effect on the p53 system, immune-dysregulation and malignant transformation were considered in the etiology of the conditions. HCV core protein may affect cancer transformation directly through an effect on a promoter gene expression. The core protein is a multifunctional protein with the capacity to bind to the so-called death domain of tumor necrosis factor receptor 1 (TNFR1) and the intracellular portion of lymphotoxin-beta receptor. The portion of TNFR1 active in apoptosis and anti apoptosis signaling pathway is the death domain affected by HCV.
5) Dermatologic manifestations are associated with treatments of HCV infection, especially interferon, e.g. IFN-induced vitiligo.

CLINICAL FEATURES

The incubation period ranges from 15–160 days (mean, 7 weeks) [3-7]. Constitutional symptoms of anorexia, nausea and vomiting, fatigue, malaise, arthralgias, myalgias, headache, photophobia, pharyngitis, cough and coryza may precede the onset of jaundice by 1–2 weeks. The nausea, vomiting and anorexia are frequently associated with alterations in olfaction and taste [3-12].

The main dermatological findings seen in most of the studies are Pruritus and prurigo, Palmar erythema, Clubbing, Hyperpigmentation, Lichen planus (Cutaneous, Oral), Leuconychia, Jaundice, Aphthous ulcers, Cutaneous vasculitis, Spider naevi, Purpura, Photosenstivity, Telangiectasia, Urticaria, Acral necrolytic erythema, Schamberg's disease, Psoriasis, Beau's lines, Porphyria cutanea tarda, Prurigo, Raynaud's phenomenon, Splinter haemorrhages, Behçet syndrome, Canities (HCV

causes sudden disruption of the melanizing function of follicles), Erythema dyschromicum perstans, any signs and symptoms of hypo- and hyperthyroidism (Immune thyroiditis is the most common extrahepatic manifestation of chronic hepatitis C infection) [3-12].

Hepatitis C is also associated with erythema nodosum, erythema multiforme, erythema induratum, autoimmune thrombocytopenia, Porokeratosis (disseminated superficial type, believed to be related to immunomodulation of the TP53 gene), Granuloma annulare, symmetric polyarthritis, scarring alopecia, Hypertrichosis of the temples, pigmentary changes, scarring, sclerodermatous changes, chloracne, ulcerations, dystrophic calcifications, sarcoidosis, Non Hodgkin's lymphoma, MALT syndrome and hepatom a [4-12].

HCV infection has been associated with several eye disorders. Keratoconjunctivitis sicca (dry eyes) is part of SS. A few cases of Mooren's ulcers have been reported. Mooren's ulcer is a rapidly progressive, painful ulceration of the cornea [5-12].

Interferon-induced dermatological diseases

Vitiligo is an autoimmune disease in which melanocytes in the skin are destroyed, with resulting depigmentation in affected areas. Although it has no specific association with liver disease, it has been linked to treatments for hepatitis C such as interferons. Interferon-induced vitiligo often completely resolves when interferon is stopped. Typical findings include aggregations of irregularly shaped white patches in a focal or segmental pattern.

Other skin conditions includes capillaritis and worsening of lichen myxedematous.

INVESTIGATIONS

Blood

Neutropenia and lymphopenia are transient and are followed by a relative lymphocytosis. Atypical lymphocytes (varying between 2 and 20%) are common during the acute phase [3,4].

Measurement of the prothrombin time (PT) is important in patients with acute viral hepatitis, for a prolonged value may reflect a severe hepatic synthetic defect, signify extensive hepatocellular necrosis and indicate a worse prognosis [3,4,6,8-12].

Blood sugar should be checked as prolonged nausea and vomiting, inadequate carbohydrate intake, and poor hepatic glycogen reserves may contribute to hypoglycaemia [3,4,6,8-12].

Stool and urine examination may be done as mild and transient steatorrhea as well as slight microscopic hematuria and minimal proteinuria are seen in some patients [3,4,6,8-12].

Liver function test

The serum aminotransferases aspartate aminotransferase (AST) and ALT (previously designated SGOT and SGPT) show a variable increase during the prodromal phase of acute viral hepatitis and precede the rise in bilirubin level. The acute level of these enzymes, however, does not correlate well with the degree of liver cell damage. A diffuse but mild elevation of the gamma globulin is common during acute viral hepatitis.

Jaundice is usually visible in the sclera or skin when the serum bilirubin value is >43 mol/L (2.5 mg/dL).

Serum alkaline phosphatase may be normal or only mildly elevated, while a fall in serum albumin is uncommon [3,4,6,8-12].

Serology

Serum IgG and IgM levels are elevated in about one-third of patients during the acute phase of viral hepatitis, but the serum IgM level is elevated more characteristically during acute hepatitis A. During the acute phase of viral hepatitis, antibodies to smooth muscle and other cell constituents may be present and low titers of rheumatoid factor, nuclear antibody and heterophil antibody can also be found occasionally. The antibodies to LKM may be positive.

A specific serologic diagnosis of hepatitis C can be made by demonstrating the presence in serum of anti-HCV which can be detected in acute hepatitis C during the initial phase. This may not be detectable in 5–10% of patients with acute hepatitis C as well as after recovery. In patients with chronic hepatitis C, anti-HCV is detectable in >95% of cases. Assays for HCV RNA by PCR is the most sensitive tests for HCV infection and represent the "gold standard" in establishing the diagnosis. However, it is not a reliable marker of disease severity or prognosis but is helpful in predicting relative responsiveness to antiviral therapy.

TREATMENT

In typical cases of acute hepatitis C, recovery is rare. Progression to chronic hepatitis is the rule and meta-analyses of small clinical trials suggest that antiviral therapy with interferon alfa monotherapy (3 million units SC three times a week) is beneficial, reducing the rate of chronicity considerably by inducing sustained responses in 30–70% of patients [13-15]. Although treatment of acute hepatitis C is recommended, the optimum regimen, duration of therapy and time to initiate therapy remain to be determined. Many authorities now opt for a 24-week course (beginning within 2–3 months after onset) of the best regimen identified for the treatment of chronic hepatitis C, long-acting pegylated interferon plus the nucleoside analogue ribavirin (1000 – 2000 mg PO), although the value of adding ribavirin has not been demonstrated [13-15].

PREVENTION

IG is ineffective in preventing hepatitis C and is no longer recommended for post exposure prophylaxis in cases of perinatal, needle stick or sexual exposure. Prevention of transfusion-associated hepatitis C has been accomplished by the following successively introduced measures: exclusion of commercial blood donors and reliance on a volunteer blood supply; screening donor blood with anti-HBc; exclusion of blood donors in high-risk groups for AIDS and the introduction of anti-HIV screening tests; and serologic and virologic screening tests for HCV infection [1,2,5-8,13-15].

In the absence of active or passive immunization, prevention of hepatitis C includes behavior changes and precautions to limit exposures to infected persons.

Anti-HCV testing is recommended for:

- anyone who received a blood transfusion or a transplanted organ before the introduction of second-generation screening tests in 1992,
- those who ever used injection drugs (or took other illicit drugs by non injection routes),
- chronically hemodialyzed patients,
- persons with clotting disorders who received clotting factors made before 1987 from pooled blood products,
- persons with elevated aminotransferase levels,
- health workers exposed to HCV-positive blood or contaminated needles,
- persons with HIV infection,
- health care and public safety personnel following a needle-stick or other non percutaneous exposure to HCV-infected material,
- sexual partners of persons with hepatitis C,
- children born to HCV-positive mothers.

For stable, monogamous sexual partners, sexual transmission of hepatitis C is unlikely and sexual barrier precautions are not necessary. For persons with multiple sexual partners or with sexually transmitted diseases, use of protection is recommended [1,2,5-8,13-15].

A person with hepatitis C should avoid sharing such items as razors, toothbrushes and nail clippers with sexual partners and family members. No special precautions are recommended for babies born to mothers with hepatitis C, and breast-feeding does not have to be restricted [1,2,5-8,13-15].

REFERENCES

1. Ali Azfar N, Zaman T, Rashid T, Jahangir M. Cutaneous manifestations in patients of hepatitis C. J Pak Associat Dermatol. 2008;18:138-43.
2. Harden MD, Skelton H, Smith KJ. Lichen planus associated with hepatitis C virus: no viral transcripts are found in the lichen planus, and effective therapy for hepatitis C virus does not clear lichen planus. J Am Acad Dermatol. 2003;49:847-52.
3. Schwartz RA, Birnkrant AP, Elmets CA, Butler DF, Callen JP, Quirk CM, et al. Cutaneous manifestations of hepatitis C. [Updated: 2013 September 12; Accessed: 2013 October 15]. Available from: http://emedicine.medscape.com/article/1134161-overview.
4. Umar M, Bushra HT, Shuaib A, Anwar A, Shah NH. Spectrum of chronic liver disease due to hepatitis C virus infection. J Coll Physicians Surg Pak. 2000;10:380-3.
5. Bovonsky HL, Mehta S. Hepatitis C a review and update. J Am Acad Dermatol. 2001;44:159-79.
6. Cacoub P, Renou C, Rosenthal E, Cohen P, Loury I, Loustaud-Ratti V, et al. Extrahepatic manifestations associated with hepatitis C virus infection. A prospective multicenter study of 321 patients. The GERMIVIC. Groupe d'Etude et de Recherche en Medecine Interne et Maladies Infectieuses sur le Virus de l'Hepatite C. Medicine (Baltimore). 2000;79: 47-56.
7. Dega H, Francès C, Dupin N, Lebre C, Simantov A, Callot C, et al. [Pruritus and the hepatitis C virus. The MULTIVIRC Unit]. Ann Dermatol Venereol. 1998;125:9-12.
8. Chuang TY, Stitle L, Brashear R, Lewis C. Hepatitis C virus and lichen planus: A case control study of 340 patients. J Am Acad Dermatol. 1999;41:787-9.
9. Tameez-ud-Deen, Naqqash S, Butt AQ. Lichen planus and hepatitis C virus infection: An epidemiologic study. J Pak Assoc Dermatol. 2003;13:127-9.
10. Ahmed I, Wahid Z, Ahmed Z. Chronic urticaria: frequency of anti-HCV antibodies. J Pak Assoc Dermatol. 2003;13:179-83.
11. Remoroza R, Bonkovsky H. Extrahepatic Manifestations of Chronic Hepatitis C. The HCV Advocate. 2003:1-3.
12. Jaeckel E, Cornberg M, Wedemeyer H, Santantonio T, Mayer J,

Zankel M, et al. Treatment of acute hepatitis C with interferon alfa-2b. N Engl J Med. 2001;345:1452-7.
13. Dienstag JL, McHutchison JG. American Gastroenterological Association technical review on the management of hepatitis C.Gastroenterology. 2006;130:231-64.
14. Ghany MG, Strader DB, Thomas DL, Seeff LB. Diagnosis, management and treatment of hepatitis C: An update. Hepatology. 2009;49:1335-74.
15. U.S. Public Health Service. Updated U.S. Public Health Service Guidelines for the Management of Occupational Exposures to HBV, HCV, and HIV and Recommendations for Postexposure Prophylaxis. MMWR Recomm Rep. 2001;50:1-52.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Clinical Image

Une tumeur infantile rare [A rare child tumour]

Salsabil Attafi, Hela Zribi

Service de Dermatologie, Hopital la Rabta, Tunis, Tunisia

Corresponding author: Dr. Salsabil Attafi, E-mail: sehlisalsabil@hotmail.com

Nous rapportons l'observation d'un nourrisson âgé de 18 mois qui avait des formations tumorales de consistance dure, rouges, douloureuses, à surface télangiectasique, siégeant au niveau des faces dorsales et palmaires du quatrième doigt droit (Fig 1).

L'examen histologique objectivait une prolifération mésenchymateuse, bénigne, fusocellulaire, de siège dermique (Figs 2 et 3).

Le diagnostic de fibromatose digitale infantile était retenu et une abstention thérapeutique était préconisée étant donné l'absence de retentissement fonctionnel.

L'évolution était marquée par la stabilisation des lésions après 6 mois de suivi.

Le fibrome digital infantile est une tumeur fibromateuse bénigne des extrémités, appartenant aux fibromatoses de l'enfant.

Il apparait dés la naissance (30% des cas) ou au cours des premiers mois de la vie sous forme de nodules dermiques uniques ou multiples, de 1 à 2 cm, durs, de couleur rose ou chair, localisés préférentiellement sur les faces d'extension des doigts et des orteils [1].

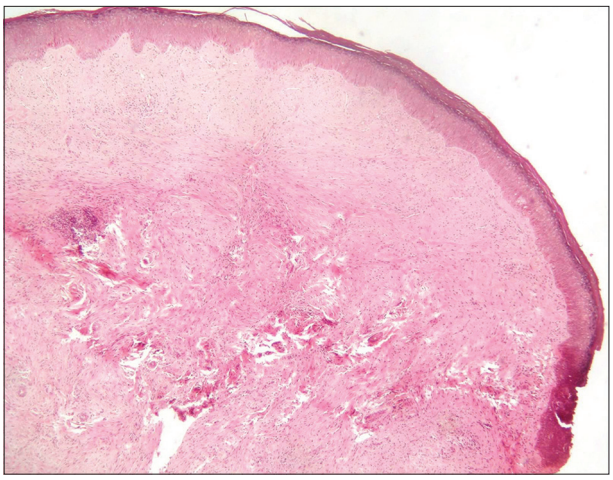


Figure 2: Prolifération fusocellulaire dermique. (HE x 10).

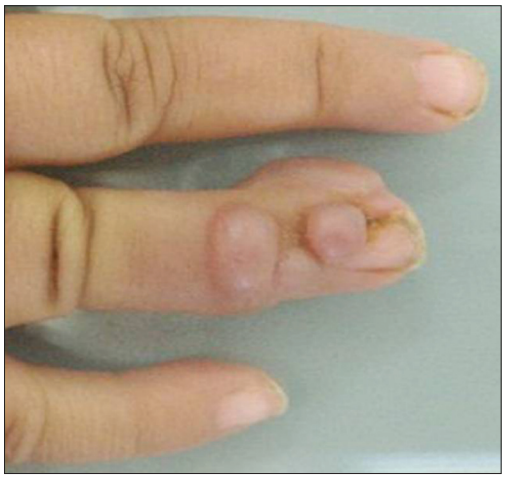


Figure 1: Nodules multiples, de couleur rose ou chair, localisés au niveau des faces dorsales et palmaires du doigt.

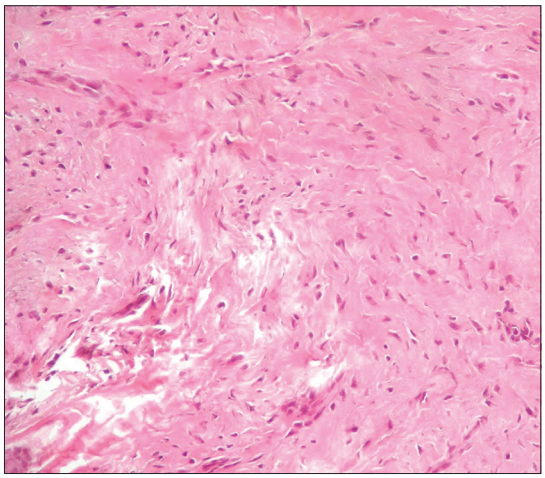


Figure 3: Cellules fusiformes régulières au sein d'un stroma collagénique. (HEx40).

How to cite this article: Attafi S, Zribi H. Une tumeur infantile rare [A rare child tumour]. Our Dermatol Online. 2015;6(4):471-472.
Submission: 10.01.2015; **Acceptance:** 16.01.2015
DOI:10.7241/ourd.20154.128

Histologiquement, on observe une prolifération dermique de fibroblastes, sans atypies cellulaires au sein d'un stroma collagénique comportant des inclusions intracytoplasmiques éosinophiles pathognomoniques.

Le diagnostic différentiel le plus fréquemment évoqué est celui de chéloïde. D'autres fibromatoses comme la fibromatose aponévrotique juvénile peuvent également être discutées [2].

Sur le plan thérapeutique, les indications d'exérèse chirurgicale doivent être limitées. L'évolution spontanée peut se faire vers la régression après quelques années. Ceci permet d'insister sur l'intérêt de temporiser devant une FDI quand elle n'a pas de retentissement fonctionnel. Cette attitude attentiste est d'autant plus justifiée que les récidives après chirurgie ne sont pas rares.

REFERENCES

1. Laskin WB, Miettinen M, Fetsch JF. Infantile digital fibroma/fibromatosis: a clinicopathologic and immunohistochemical study of 69 tumors from 57 patients with long-term follow-up. Am J Surg Pathol. 2009;33:1-13.
2. Niamba P, Léauté-Labrèze C, Boralevi F, Lepreux S, Chamaillard M, Vergnes P, et al. Further documentation of spontaneous regression of infantile digital fibromatosis. Pediatr Dermatol. 2007;24:280-4.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Clinical Image

Facial nevus spilus mistakenly treated as melasma

Tasleem Arif, Syed Suhail Amin

Postgraduate, Department of Dermatology, STDs and Leprosy, Jawaharlal Nehru Medical/College(JNMC), Aligarh Muslim University (AMU), Aligarh, India.

Corresponding author: Dr. Tasleem Arif, E-mail: dr_tasleem_arif@yahoo.com

A 17 year old male visited our dermatology outpatient department (OPD) with brownish hyperpigmentation over left side of the face. The patient had visited some physicians in the past for this hyperpigmentation and had been applying several demelanizing creams but with poor response to the treatment. On further enquiry, he revealed that this pigmentation has been since his childhood. There is no history of intensification of this pigmentation during summers. On examination, there is a large macule around the size of 7cm×5cm, slightly brownish in color involving major portion of the left check and submandibular area with serrated borders on the superior and medial sides of the macule (Fig. 1a). On this light brownish macular background, there are present multiple 1-2 mm dark brownish macules along with some darker brownish papular lesions (nevi). There was no pigmentation on the right side of the face (Fig. 1b). With such a history and further supported by cutaneous examination, a diagnosis of unilateral facial nevus spilus was made.

Nevus spilus is also called as Speckled and lentiginous nevus. It is regarded as congenital melanocytic nevus. It remains usually lentiginous in early childhood and may develop palpable components at puberty in a 'speckled' distribution. It is composed of a flat, macular component which is slightly darker than the surrounding normal skin. There may be lentigo-like lesions or elevated darker-brown nevi interspersed within the slightly brownish macular background [1].

The present case was the nevus spilus which has been wrongly treated as melasma. Many general physicians treat any facial pigmentation as melasma with over the counter demelanizing creams especially steroid containing triple combinations. However, applying the basic principle of thorough history taking and cutaneous examination can avoid such mistakes.

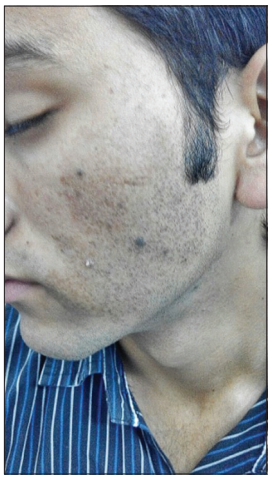


Figure 1a: Speckled and lentiginous nevus on left side of the face in a 17 year male.

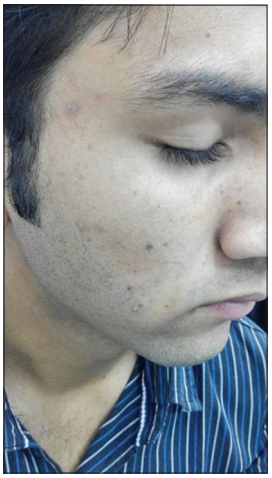


Figure 1b: Same patient with normal right side of the face.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

How to cite this article: Arif T, Amin SS. Facial nevus spilus mistakenly treated as melasma. Our Dermatol Online. 2015;6(4):473-474.
Submission: 30.03.2015; **Acceptance:** 04.06.2015
DOI: 10.7241/ourd.20154.129

Written informed consent was obtained from the patient for publication of this article and any accompanying images.

REFERENCES

1. Newton Bishop JA. Lentigos, Melanocytic Naevi and Melanoma. In: Burns T, Breathnach S, Cox N, Griffiths C. Eds. Rook's Textbook of Dermatology. Wiley-Blackwell, U.K, 8th edition 2010; 3: 54.14-54.15.

Source of Support: Nil, **Conflict of Interest:** None declared.

Letter to the Editor

Did Sushrutha first describe ear lobe repairs? A peep into the Samhita

Harinatha Sreekar[1], Ravi Reddy[2], Nithya Raghunath[3], Nikhitha Raghunath[4], Harinatha Sreeharsha[5]

[1]Department of General Surgery, MVJ Medical College, Bangalore, Karnataka, India, [2]Department of General Surgery, Vijaynagar Institute of Medical Sciences, Bellary, India, [3]Contura Cosmetic Clinic and Department of Dermatology, MVJ Medical College, Bangalore, Karnataka, India, [4]Department of Community Dentistry, Oxford Dental College, Bangalore, Karnataka, India, [5]Department of Urology, Fortis Hospital, Bangalore, Karnataka, India

Corresponding author: Ass. Prof. Harinatha Sreekar, E-mail: drsreekarh@yahoo.com

Sir,

Ear lobe repairs are among the commonest and simplest procedures performed by plastic surgeons in India. The heavy jewelry that's part of the Indian tradition surely is the culprit. Given the prevalence of the elongated (or torn) ear lobes in India; it's only logical that surgeons of yore ventured into remedying this simple issue. Now a days there are many methods of repairing a partial or completely torn ear lobe [1,2]. The techniques have ranged from simple adjustment of local skin to more imaginative sandwiching of conchal cartilage [3]. Who first described the ear lobe repair however is unclear.

Sushrutha, now widely regarded as the 'father of plastic surgery' in his treatise 'Sushrutha Samhita' seems to describe in fairly elaborate detail the method of repairing a torn ear lobe. He even described over fifteen different procedures based on the nature and size of ear lobe tear. Some include

1. The Nemi- Bandhinaka: To be used in cases where the bifurcated lobes and equal in size.
2. The Utpala-Bhedyaka: To be used in cases where the severed lobes are round, extended, and equal.
3. The Valluraka: To be used in cases where the severed lobes of the ears would are short, circular and equal in size.
4. The Aangima: To be adopted in cases where the anterior surface of the torn lobe is more elongated than the other.
5. The Ganda-Karna: To be adopted in cases where the posterior surface of the torn lobe is more elongated than the other.

Though the exact details of the surgical nuances are difficult to decipher, he does mention some valid and important aspects of lobe repair. He mentions slicing off a patch of healthy skin from the cheeks and adhering it to one of the severed lobes (Akin to a skin graft). He describes a process called the Kapata-Sandhika (closing of the two leaves of a door {Kapatam}) wherein he brings about an adhesion, on the posterior side, between the bifurcated lobes and another, which by shortening the elongated anterior side of the ear (Akin to a wedge excision or a Z-plasty). He also describes the Ardha-Kapata-Sandhika (Ardha{Half} Kapata {Door} Sandhika {Joining}) [4]. This description has a peculiar resemblance to the half Z-plasty. The nature of Sushrutha's contribution to reconstructive surgery is a matter of controversy which is unlikely to die down in the near future. Part reason for such confusion is the presence of numerous translations and even more numerous interpretations. However, the very fact that he might have described procedures like adhesion, Z-plasty and half Z-plasty is a matter of awe in itself.

REFERENCES

1. Zilinsky I, Tessone A, Winkler E, Mendes D, Liran A. Partially torn ear lobe repair using a cross-stitching technique. Dermatol Surg. 2009;35:987-9.
2. Reiter D, Alford EL. Torn earlobe: a new approach to management with a review of 68 cases. Ann Otol Rhinol Laryngol. 1994;103:879-84.

How to cite this article: Sreekar H, Reddy R, Raghunath N, Raghunath N, Sreeharsha H. Did Sushrutha first describe ear lobe repairs? A peep into the Samhita. Our Dermatol Online. 2015;6(4):475-476.

Submission: 01.02.2015; **Acceptance:** 10.07.2015

DOI:10.7241/ourd.20154.130

3. Agarwal R, Chandra R. A new technique for repair of acquired split-ear-lobe deformity: the free conchal cartilage sandwich graft, J Plast Reconstr Aesthet Surg. 2010;63:499-505.
4. Kunja Lal Bhishagratna K. An English translation of the Sushruta samhita, based on original Sanskrit text: JN Bose College Square, Calcutta,1907:143-7.

Source of Support: Nil, **Conflict of Interest:** None declared.

Letter to the Editor

Cement burn

Natsuko Matsumura, Masato Ishikawa, Tomoko Hiraiwa, Nobuyuki Kikuchi, Yasunobu Kato, Toshiyuki Yamamoto

Department of Dermatology, Fukushima Medical University, Fukushima, Japan

Corresponding author: Dr. Natsuko Matsumura, E-mail: natsukom@fmu.ac.jp

Sir,

A 22-year-old Japanese man was admitted to our emergency department with sharp pain to both his hands and wrists due to chemical burns sustained while working at a construction site. Cement had accidentally dripped down his gloves and made contact with his hands and wrists. Five hours later, he felt a burning sensation and noticed blisters at the affected regions. The symptoms progressively worsened and became increasingly painful and swollen. Physical examination revealed erythematous, edematous lesions and ulcers with minimal necrosis (Fig. 1a and b). His vital signs were within normal range, and no abnormality was detected on the rest of his physical examination. Except for highly elevated creatinine kinase (631 U/L; normal range 62-287 U/L), laboratory findings, including complete blood count, liver and renal function test, and serum electrolytes, were normal. Initial treatment consisted of debridement and analgesia and the ulcers were epithelized by conservational therapy within 2 weeks. Four weeks later, he visited our department and presented with only mild contracture on the surface of his wrists. A patch test, which was performed to investigate whether underlying contact allergies to cement existed, was uniformly negative.

Cement is widely used in the construction sector, and contact with the substance occasionally induces several types of cutaneous reactions. The most common cement-related rash is allergic contact dermatitis, especially that due to chromate and cobalt. Irritant reactions are also common and usually mild. By contrast, cement burns lead to severe symptoms that require intensive therapy [1].

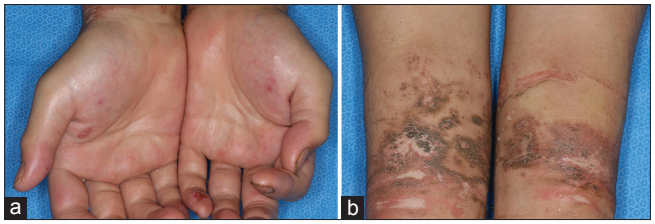


Figure 1: Clinical appearance of hands (a) and wrists (b) several hours after contact with wet cement.

Cement burns are caused by alkaline calcium hydroxide, which is formed when water is added to cement. Cement burns are thought to be developed by direct contact of cement to skin. Several etiological factors, such as prolonged contact with wet, strongly alkaline cement, the abrasive effect of prolonged rubbing of clothing, boots or gloves impregnated with alkali, as well as aggregates of cement, may be harmful. The pH range of wet cement is 10-12 but can reach 12-14 during the process stage [1,2].

Chemical burns from cement cause erythema, bullae, and pain within a few hours after contact, and result in acute ulcerative dermatitis. Such burns are insidious in onset, and patients may therefore only initially be aware of minor irritation. However, this progressive tendency of cement burns often leads to full-thickness burns that require skin grafting [3].

Because of the progressive nature and severity of the tissue destruction caused by cement, prompt removal of contaminated clothing and thorough washing of the skin with water is advised [3]. Adequate hazard notifications and information are especially important in avoiding such accidents [4].

How to cite this article: Matsumura N, Ishikawa M, Hiraiwa T, Kikuchi N, Kato Y, Yamamoto T. Cement burn. Our Dermatol Online. 2015;6(4):477-478.
Submission: 11.03.2015; **Acceptance:** 18.07.2015
DOI:10.7241/ourd.20153.131

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Mehta R K, Handfield-Jones S, Bracegirdle J, Hall PN. Cement dermatitis and chemical burns. Clin Exp Dermatol. 2002;27:347-8.
2. Avnstrop C, Carmody M. The dermal toxicity of cement. Toxicol Ind Health. 2002;18:321-31.
3. Keles A, Aygencel G, Kahveci O, Bildik F, Demircan A. Contact with wet cement: report of a case. Contact Dermatitis. 2008;58:173-4.
4. Spoo J, Elsner P. Cement burns: a review 1960-2000 Contact Dermatitis. 2001;45:68-71.

Source of Support: Nil, **Conflict of Interest:** None declared.

Letter to the Editor

Pleomorphic basal cell carcinoma: report of an uncommon histological variant

Mariem Bel Haj Salah, Anissa Zaouek, Ines Smichi, Wafa Koubâa, Achraf Chadly-Debbiche

Department of Pathology, Habib Thameur Hospital, Tunis, Tunisia

Corresponding author: Dr. Ines Smichi, E-mail: smichi.ines@yahoo.fr

Sir,

Basal cell carcinoma (BCC) is the most common malignant skin tumor. It forms different histologic patterns that often have variable outcomes and prognoses. Pleomorphic BCC is an uncommon pathologic variant of unknown pathogenesis. Herein, we report a case of pleomorphic BCC in a patient with Xeroderma Pigmentosum (XP).

A 42-year-old male with a medical history of XP consulted for a nodular, pigmented tumor of the temporal region measuring 5cm in its greatest dimension. Carcinologic surgical removal of the tumor was performed. At histopathologic examination, it was composed of nodular aggregates of atypical basaloid cells with peripheral palisading and stromal retraction. In addition, distributed throughout the tumor nests, there were numerous giant cells [Fig. 1]. These cells had an ill defined abundant eosinophilic cytoplasm and multiple large, irregular, hyperchromatic nuclei. Numerous abnormal mitotic figures were also observed [Fig. 2]. The overall histologic features were highly suggestive of pleomorphic BCC. The patient was regularly followed in the dermatology department with no recurrence after an eight month follow-up.

Pleomorphic BCC have been variously reported as BCC with pleomorphic giant cells, basal cell epithelioma with monster cells and basal cell epithelioma with giant tumor cells [1-4]. It is an exceedingly rare variant of BCC with less than 60 cases reported in the literature [5]. According to a large study of 52 cases of pleomorphic BCC, it accounts for approximately 1 to 2,5% of all BCC [6]. It usually affects elderly adults with an average age of 65 years old and no sex predominance. Our case is original by the age of onset (42 years old); this may be explained by the fact that our patient has XP.

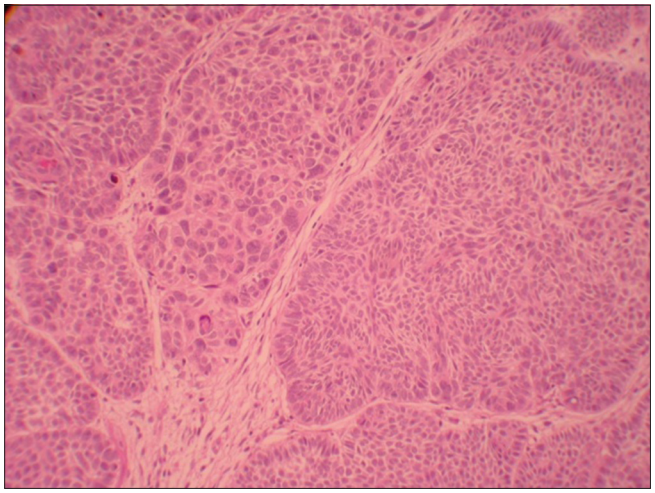


Figure 1: Pleomorphic basal cell carcinoma. Enlarged nuclei and giant cells are present within the lobules of nodular basal cell carcinoma. (HE X 100).

Clinically, pleomorphic BCC usually exhibits a typical nodular appearance, as in our patient, and has a propensity for the head and neck region [7]. Histologically, tumor is usually well circumscribed and solid but occasionally with adenoid or cystic features. The cardinal sign is the presence of enlarged mononuclear and/or multinucleated tumor cells scattered throughout the lobules. These cells have hyperchromatic and irregularly outlined nuclei with a vesicular appearance. Prominent nucleoli are also occasionally seen. Mitoses are often raised but are not necessarily atypical as in our case [4].

How to cite this article: Bel Haj Salah M, Zaouek A, Smichi I, Koubâa W, Chadly-Debbiche A. Pleomorphic basal cell carcinoma: report of an uncommon histological variant. Our Dermatol Online. 2015;6(4):479-480.

Submission: 16.02.2015; **Acceptance:** 21.08.2015

DOI:10.7241/ourd.20154.132

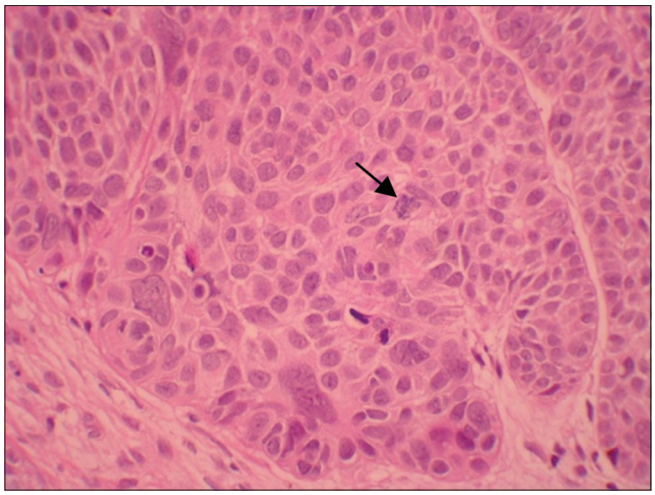


Figure 2: Bizarre cells have multiple, sharply nucleolated nuclei. Note the abnormal mitosis (arrow). (HE X 400).

In all cases tested, aneuploidy was identified in these bizarre cells [4,7]. Enhanced expression of proliferation-associated antigens (PCNA, Ki-67) has also been reported in these enlarged cells [1,4] as well as positivity of Bcl-2. This immunoprofile indicates that the giant cells are cycling and do not appear to represent a senescent change [8].

In one report of this variant, similar giant cells were found in the surrounding stroma. They were believed to derive from the same tissue lineage as the pleomorphic cells within tumor nodules [4].

The presence of atypical giant cells doesn't darken in any case the prognosis which is similar to that of the classical form [5]. Management of these tumors usually consists in a wide local excision with usually a favorable outcome and no recurrences as in our patient.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Cultan RT, Maluf HM. Immunohistochemical characterization of pleopmorphic giant cells in basal cell carcinoma. J Cutan Pathol. 1999;26:353-6.
2. Elston DM, Bergfeld WF, Petroff N. Basal cell carcinoma with monster cells. J Cutan Pathol. 1993;20:70-3.
3. Ono T, Egawa K, Higo G Fallas VH. Basal cell epithelioma with giant tumor cells. J Dermatol. 1985;12:344-8.
4. Alan S. Boyd. Tumors of the Epidermis; In Dermatopathology (Raymond L Barnhill, Arthur Neil Crowson, Cynthia M. Magro, Michael W. Piepkorn, eds), 3rd edn. McGraw-Hill Medical. 2010 :556-614.
5. Huang CC. Pleomorphic Basal cell carcinoma. South Med J. 2006;99:200.
6. Tschen JP, Cohen PR, Schulze KE, Tschen JA, Nelson BR. Pleomorphic basal cell carcinoma: case reports and review. South Med J. 2006;99:296-302.
7. Garcia JA, Cohen PR, Herzberg AJ, Wallis ME, Rapini RP. Pleomorphic basal cell carcinoma. J Am Acad Dermatol. 1995;32:740.
8. Jakobiec FA, Zakka FR, Townsend DJ. Pleomorphic basal cell carcinoma of the eyelid with true ductular differentiation. Graefes Arch Clin Exp Ophthalmol. 2012;250:451-4.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Letter to the Editor

Entodermoscope: A tool to diagnose and monitor pediculosis captitis

Shetty Shricharith, Jindal Anuradha, Rao Raghavendra, Sathish Pai

Department of Dermatology, Kasturba Medical College, Manipal University, Manipal, Karnataka, India

Corresponding author: Dr. Shetty Shricharith, E-mail: drshricharith@gmail.com

Sir,

Head lice infestation, or pediculosis capitis, is a common health concern, especially in children. Diagnosis is based upon the detection of lice or nits. However, because the louse moves quickly and avoids light, it's often invisible to the naked eye. Nits containing vital nymphs may look similar to empty nits and so-called pseudo-nits, such as hair casts, debris, scales from seborrheic dermatitis to naked eye [1]. Dermoscopy can aid in overcoming these diagnostic hurdles. We here report a case of 12-year- old girl who was referred for the complaint of chronic itching of scalp and neck area since last 7months. Physical examination revealed the presence of excoriated lesions at the occipital and cervical regions along with numerous yellow to brown nits, fixed to hair shaft. Dermoscopic examination using non-contact polarized light dermoscope (Dermlite; 3Gen, USA) along with Nikon D3200 digital camera, revealed the presence of multiple yellow to brown, ovoid eggs firmly attached to the hair shaft, corresponding to nits with vital nymph, along with brown translucent flat empty nits (Fig. 1). On further examination of occipital area of scalp, we were able to see multiple live lice (Pediculus humanus var. capitis) (Fig. 2). A diagnosis of pediculosis capitis was made and she was advised topical 1% permethrin lotion and oral ivermectin as per standard protocol along with nit combing. After first week, dermoscopy revealed persistence of brown ovoid nits and patient was asked to continue the treatment. After 3 weeks of treatment patient reported improvement and dermoscopy revealed no nits or lice.

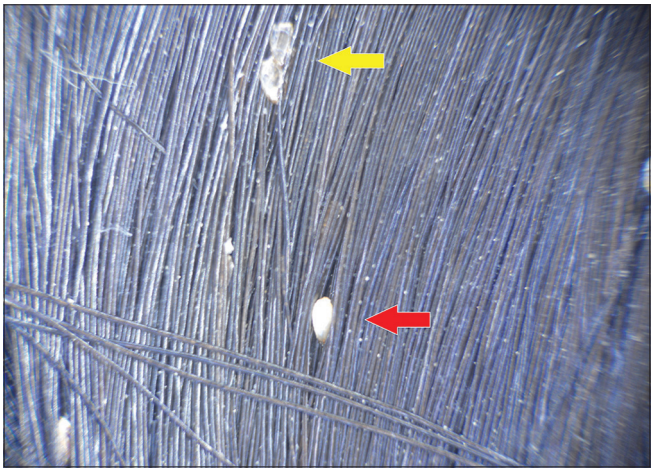


Figure 1: Translucent empty nit case (yellow arrow) and opaque ovoid nit case with vital nymph (red arrow).

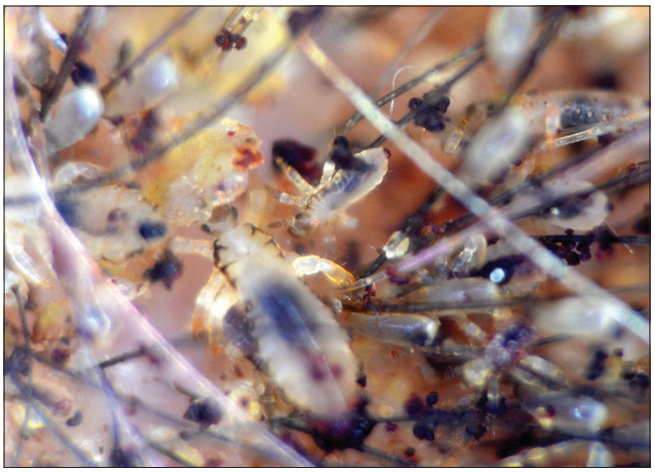


Figure 2: Lice attached to hair shaft near to scalp surface.

Dermoscopy is a non-invasive technique that allows a rapid and magnified in vivo observation of the skin with the visualization of morphologic features invisible to the naked eye [2]. Although dermoscopy was initially developed for the diagnosis of pigmented lesions, it has been used as an aid to diagnosis in squamous diseases,

How to cite this article: Shricharith S, Anuradha J, Raghavendra R, Pai S. Entodermoscope: A tool to diagnose and monitor pediculosis captitis. Our Dermatol Online. 2015;6(4):481-482.

Submission: 01.03.2015; **Acceptance:** 05.05.2015

DOI:10.7241/ourd.20153.133

depigmenting diseases, infections and infestations [3]. The term "entodermoscopy" was coined to refer to the use of dermoscopy as an aid in the diagnosis and follow-up of treatment of infestations such as scabies, pediculosis, tungiasis, cutaneous larva migrans and tick infestations [3,4]. New generation non-contact dermoscope using polarized light prevents the possible risk of transfection in the latter cases. Dermoscope hence not only provides an easy way to establish confirmed diagnosis but also help in monitoring the disease after starting the treatment, especially in the era where resistance to pediculicides becoming an emerging problem in many parts of the world.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Di Stefani A, Hofmann-Wellenhof R, Zalaudek I. Dermoscopy for diagnosis and treatment monitoring of pediculosis capitis. J Am Acad Dermatol. 2006;54:909-11.
2. Micali G, Tedeschi A, West DP, Dinotta F, Lacarrubba F. The use of videodermatoscopy to monitor treatment of scabies and pediculosis. J Dermatolog Treat. 2011;22:133-37.
3. Zalaudek I, Giacomel J, Cabo H, Di Stefani A, Ferrara G, Hofmann-Wellenhof R, et al. Entodermoscopy: a new tool for diagnosing skin infections and infestations. Dermatology. 2008;216:14–23.
4. Tschandl P, Argenziano G, Bakos R, Gourhant JY, Hofmann-Wellenhof R, Kittler H, et al. Dermoscopy and entomology (entomodermoscopy). J Dtsch Dermatol Ges. 2009;7:589–96.

Source of Support: Nil, **Conflict of Interest:** None declared.

Letter to the Editor

A case of inverse psoriasis with interdigital involvement

Müzeyyen Gönül[1], Hasan Benar[1], Aysun Gökce[2], Murat Alper[2]

[1]Dermatology Clinic, Dışkapı Yıldırım Beyazıt Education and Research Hospital, Ankara, Turkey, [2]Pathology Clinic, Dışkapı Yıldırım Beyazıt Education and Research Hospital, Ankara, Turkey

Corresponding author: Assoc. Prof. Müzeyyen Gönül, MD, E-mail: muzeyyengonul@gmail.com

Sir,

Inverse psoriasis is an uncommon form of psoriasis that involves intertriginous areas such as axillae and inguinal creases [1]. Interdigital psoriasis (IP), a subtype of inverse psoriasis, has been defined firstly as a distinct entity by Waisman in 1961 and this entity was called as "white psoriasis" or "psoriasis alba" [2]. There are few reported cases of IP in literature [2-4]. This report presents a case of IP who has been misdiagnosed as tinea pedis for one year and then diagnosed with the development of psoriasis lesions in other intertriginous areas.

A 65-year-old woman attended with the complaints of erythematous pruritic eruptions on his axillae, inframammary and inguinal regions. The lesions had appeared on the toe web one year ago and they did not change although the patient took topical antifungal therapies several times. Later, complaints of pruritus and dandruff of scalp was added. 15 days ago red pruritic rash occured on her axillae, inguinal creases and inframamary areas. Grouped, erythematous and squamous papules on the axillae, inguinal and inframamary areas and bilaterally whitish plaque and desquamation on the 4. and 5.web of toes were detected on dermatological examination (Fig. 1). There were distal onycholysis, subungual hyperkeratosis and melanonychia striata on the toe-nails. Wood lamp examination and KOH preparation were negative. Histopatological examination of biopsy material obtained from interdigital area showed focal parakeratosis, thinning of granular layer, regular acanthosis and perivascular infiltration (Figs 2 a and b). Inverse psoriasis with interdigital involvement was diagnosed based on clinical and histopatological findings. Topical corticosteroid therapy was suggested and the complaints of the patient improved significantly.

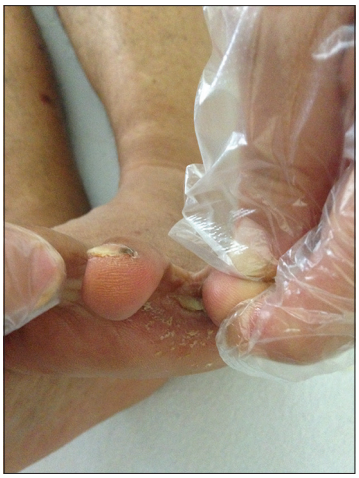


Figure 1: Whitish plaque on the web of toes.

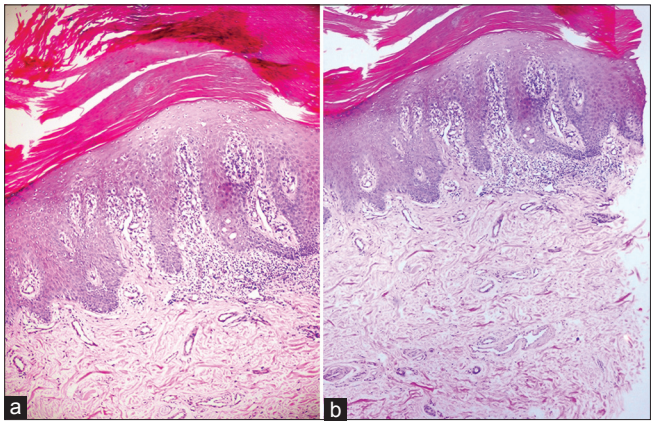


Figure 2: a) Focal parakeratozis, thinning of granular layer, regular acathosis (H&Ex40) b) Tendency of coalescence of rete ridges and perivascular mononuclear cells infiltration on the superficial dermis (H&Ex100).

The patient's informed consent was obtained.
Prior to the study, patient gave written consent to the examination and biopsy after having been informed about the procedure.

How to cite this article: Gönül M, Benar H, Gökce A, Alper M. A case of inverse psoriasis with interdigital involvement. Our Dermatol Online. 2015;6(4):483-484.
Submission: 13.03.2015; **Acceptance:** 05.05.2015
DOI:10.7241/ourd.20153.134

IP involves the intertoe spaces of one or both feet and is clinically characterized with macerated white, sodden patches/plaques resembling interdigital fungal infections [2-4]. There is little or no itching but sometimes fissures may occur and then itching may become more severe. The present case had severe itching [4].

The cases of IP may have other stigmatas of psoriasis and histopathological findings of IP resemble classical psoriasis but varying degrees of alterations such as atypical or incomplete parakeratosis and intermittant or intact stratum granulosum may be seen [4]. In our case, incomplete parakeratosis was seen.

Nowadays, it is debatable if IP is a distinct entity or not. Although some authors suggest that IP is a distinct atypical form of psoriasis, in a recent study, it is reported that IP is not a distinct form and it may be seen in 3.66% of the modarete or severe psoriasis patients [2-4].

IP is clinically important as it is often misdiagnosed and commonly mistaken for interdigital fungal infections [2-4]. Sometimes, coexistence of IP and interdigital fungal infections may occur, while fungal infections may superimpose psoriasis lesions, psoriasis lesions may be triggered by fungal infections due to Koebner phenomenon [4].

The treatment of IP is similar to treatment of inverse psoriasis but it may be resistant to therapy and may shows recurrences [2].

Consequently IP is a misdiagnosed form of psoriasis because of clinical similarity to fungal infections. Diagnosis of this entity may be more difficult if it occurs earlier than psoriasis lesions as in our case. So, IP must be kept in mind in patients who have interdigital lesions particularly unresponsive antifungal therapies. If native examination and fungal culture are negative skin biopsy must be obtained from interdigital lesions. These simple methods supply certain diagnosis and prevent the use of unnecessary drugs.

CONSENT

The examination of the patient was conducted according to the Declaration of Helsinki principles.

REFERENCES

1. Syed ZU, Khachemoune A. Inverse psoriasis: case presentation and review. Am J Clin Dermatol. 2011;12:143-6.
2. Waisman M. Interdigital psoriasis ("white psoriasis"). Arch Dermatol. 1961;84:733-40.
3. Mommers JM, Seyger MM, van der Vleuten CJ, van de Kerkhof PC. Interdigital psoriasis (psoriasis alba): renewed attention for a neglected disorder. J Am Acad Dermatol. 2004;51:317-8.
4. Bardazzi F, Antonucci VA, Patrizi A, Alessandrini A, Tengattini V, et al. Interdigital psoriasis of the feet (psoriasis alba): not a distinct form of psoriasis. Dermatology; 2013;227:130-3.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Letter to the Editor

General public perception of a dermatologist in urban India

Ravi Reddy[1], Harinatha Sreekar[2], Nithya Raghunath[3], Harinatha Sreeharsha[4]

[1]Department of General Surgery, Vijaynagar Institute of Medical Sciences, Bellary, India, [2]Department of General Surgery, MVJ Medical College, Bangalore, Karnataka, India, [3]Contura Cosmetic Clinic and Department of Dermatology, MVJ Medical College, Bangalore, Karnataka, India, [4]Department of Urology, Fortis Hospital, Bangalore, Karnataka, India

Corresponding author: Ass. Prof. Sreekar Harinatha, E-mail: drsreekarh@yahoo.com

Sir,

Dermatology and its practitioners has always been considered a niche segment. Though they cater to a variety of disorders ranging from debilitating skin diseases to cosmetology, the public perception though could not be different. To analyse the perception a bit more objectively, we conducted a short survey (Using www.surveymonkey.com) among the general public. We surveyed a total of 386 respondents. We focussed on identifying their perception and the importance given to dermatologists in comparison to other fields of medicine.

Some of the significant findings are as below:

1. 62% perceived that dermatologists cater primarily to cosmetic issues
2. 28% perceived that dermatologists treat skin cancer primarily
3. 83% opined that dermatology is of lesser significance compared to General surgery, Medicine, Gynecology and Pediatrics.
4. 76% believed that dermatologists need to work less harder than their peers in other fields
5. 82% believed that dermatologists make more money than others
6. 17% opined that dermatologists can treat life threatening disorders
7. 53% believed that practicing dermatology is easier compared to others in terms of patient expectations and stress

While most of the findings are exceptionally skewed, it however reflects the state of perception of general public in urban India towards dermatologists. Such findings have been replicated in similar studies in different settings [1,2]. This raises concern towards not only the image projected by the medical fraternity but also the role played by the media. Media tends to brand dermatologist as 'Cosmetologists' thus significantly dwarfing the status in the eyes of the public. This calls for creating more awareness in the general public with regards to the varying aspects and expertise needed and consistently expected from a dermatologist.

REFERENCES

1. Brezinski EA, Harskamp CT, Ledo L, Armstrong AW. Public perception of dermatologists and comparison with other medical specialties: Results from a national survey. J Am Acad Dermatol. 2014;71:875-81.
2. Schwartzbaum AM, McGrath JH, Rothman RA. The perception of prestige differences among medical subspecialties. Soc Sci Med. 1973;7:365–71.

Source of Support: Nil, **Conflict of Interest:** None declared.

How to cite this article: Reddy R, Sreekar H, Raghunath N, Sreeharsha H. Generalized keratosis pilaris rubra. Our Dermatol Online. 2015;6(4):485-485.
Submission: 03.02.2015; **Acceptance:** 09.07.2015
DOI:10.7241/ourd.20154.135

Our Dermatology Online

Historical Article

Jean Alfred Fournier (1832-1914): His contributions to dermatology

Nadeem Toodayan

Associate Lecturer, University of Queensland School of Medicine, Southern Clinical School, Brisbane, Australia

Corresponding author: Dr. Nadeem Toodayan, E-mail: nadeem_toodayan@hotmail.co.uk

INTRODUCTION

Historically, the interplay between dermatologists and other specialists has nowhere been more pronounced than in the field of venereology. A true sister to the art, it has accurately been remarked that *dermatology and venereology were inextricably intertwined* all throughout the nineteenth century [1]. At that time, the range of figures thus inclined were themselves a motley bunch, ranging in focus from the pure dermatologist – of which there were many – to medical polymaths like Sir Jonathan Hutchinson (1828-1913), who excelled in both dermatology and venereology, as well as in everything else he pursued. In amongst this peculiar group, we here remember Jean Alfred Fournier (1832-1914), the obligate venereologist, who, out of this innate overlap, forged a most fruitful and memorable career path. Fournier's contributions to modern dermatology are numbered but his influence on the specialty as the first ever Professor of syphilology and cutaneous diseases in Paris should not be underestimated. Written in recognition of the recently passed centennial anniversary of his death – 25th December 2014 –, this article will serve to remind readers of Fournier's legacy by reviewing some of his specific contributions to dermatology.

FOURNIER'S GANGRENE

When spoken of today, the likely vestige of Alfred Fournier's legacy most often inciting further discussion about the man, is the fulminating genital gangrene described by him in the early 1880s and commonly referred to by his name. Doubtless the most popular and widely known of the Fournier eponyms, Fournier's gangrene certainly falls within the range of skin disease, and a brief dermatologic account of the condition will therefore serve well to reintroduce Alfred Fournier to the reader (Fig. 1).

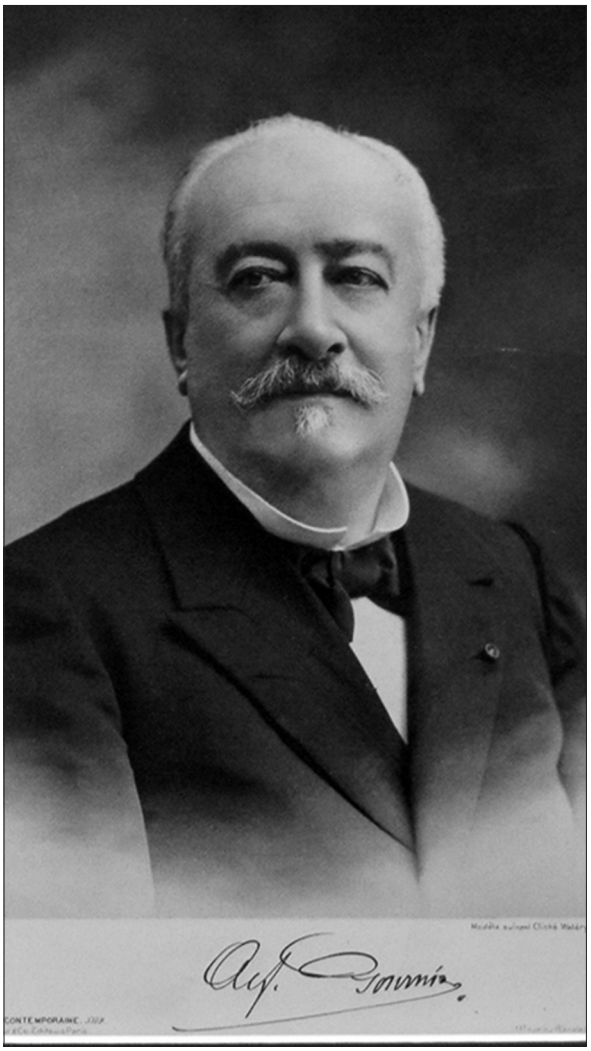


Figure 1: Professor Jean Alfred Fournier (1832-1914); the archetypal dermatovenereologist. Reproduced from an original photo-print owned by the author.

How to cite this article: Toodayan N. Jean Alfred Fournier (1832-1914): His contributions to dermatology. Our Dermatol Online. 2015;6(4):486-491.
Submission: 31.05.2015; **Acceptance:** 16.07.2015
DOI: 10.7241/ourd.20154.137

The peculiar anatomy of the human perineum means that gangrene affecting the genitalia is often a rapidly progressive and life threatening condition. Typically induced by soft tissue damage in the immunocompromised patient – diabetics and alcoholics are classical candidates – Fournier's gangrene is best described as a fulminating and polymicrobial necrotising fasciitis of the genitalia [2]. Generally speaking, it is more common in men but can occur in women and even children. Synergistic infection with aerobic and anaerobic organisms in the setting of a toxin mediated obliterative endarteritis is primarily responsible for this type of gangrene, and by spreading along the deep fascial planes, the infective cascade is rarely inhibited in its extension. Cutaneous infection precedes genital gangrene in up to 21% of cases. In these patients, poor genital hygiene, pressure sores, skin trauma from any cause, and hidradenitis suppurativa present common predisposing factors. Surgical debridement and broad spectrum antibiosis with supportive care remains the mainstay of therapy in patients with Fournier's gangrene.

The clinical picture of Fournier's gangrene has often been told by the dermatologist, not least so because many of the initial and final features of the disease are cutaneously manifested. What starts out as a severely painful erythematous swelling of the genitalia, often progresses to a less irritating and finally painless area of genital gangrene, the characteristic dark purple or black discolouration of which, is typical of gangrenous changes occurring anywhere else. Fever, malaise, lethargy, and other signs of systemic toxicity occur early on and are commonly associated features. Specific skin signs to exclude when assessing for Fournier's gangrene include palpable cutaneous crepitus, small erupting vesicles, oedema exceeding the area of erythema, induration, cyanosis, bronzing, and blistering of the skin, and dermal thrombosis [3,4]. A lack of lymphangitis in the presence of these features may also be suggestive of deeper skin infection. When in doubt of the diagnosis, certain dermatologic differentials to have in mind include severe scrotal cellulitis, ecthyma gangrenosum, vasculitis associated genital gangrene, warfarin induced skin necrosis, and genital pyoderma gangrenosum [5]. The erosive and gangrenous balanitis of Corbus [6], which is classically brought on by anaerobic organisms in the uncircumcised patient and usually limited to the glans penis only, is likewise to be distinguished from Fournier's gangrene.

'GANGRENE FOUDRAYANTE DE LA VERGE'

Alfred Fournier first lectured on genital gangrene in December of 1883, at which time he focused on delineating what he called *gangrene foudroyante de la verge* – or fulminating gangrene of the penis – from what he felt were other less aggressive types of genital gangrene [7]. Having seen five such cases to date, he felt it important to delegate this type of gangrene a distinct seat in the medical nosology, and on introducing the specific terminology above mentioned – *foudroyante* – he succeeded in doing just that. Fournier noted diabetes, alcoholism, and vascular insufficiency as important constitutional predisposing factors in many instances, but these cases were to be separated from what he classed as 'fulminating' genital gangrene. He concluded his first lecture asserting that *there exists a different gangrene of the penis from the other types involving the same organ*, and presented a pointed list emphasizing its most unique characteristics: *the absence of any predetermined cause *, a gangrenous and sudden beginning, astonishingly rapid and always considerable extension, the frequent coexistence of purple discoloration, and finally an excessive morbidity*. The clinical features and natural history of the disease were masterly detailed in a follow up lecture given in February of 1884 [8]. Noting the disease's malignant potential, shocking speed of invasion, systemic features, and spontaneous occurrence in what were otherwise apparently healthy young males, Fournier concluded that it was likely infectious in nature. He also highlighted the strong potential for mortality in these patients.

Fournier's lectures on genital gangrene hardly occupied a significant place in his outstanding oeuvre, and his enquiries into the subject would almost have been merely a side effect of his intensive interests in venereology. That being said, there is no taking away from the influence of his lectures, as both were, in the manner of everything else he studied, role models of clinical excellence. We have from perusing them, a very high estimate of the standard quality of his daily works, and the eponym Fournier's gangrene preserves prestige in having conveyed to us, the wonderful legacy of one of France's finest clinicians.

*Improvements in our understanding of the pathophysiological processes occurring in necrotising fasciitis means that modern medicine recognizes no such thing as truly 'idiopathic' genital gangrene.

Even so, the rest of Alfred Fournier's classical clinical description remains highly accurate, and the name Fournier's gangrene is today used to refer to rapidly fulminating genital gangrene of any aetiology.

CLINICAL CONTRIBUTIONS TO DERMATOLOGY

Alfred Fournier's phenomenal clinical ability was largely built upon meticulously describing the signs and symptoms of syphilis, a disease with which his name was almost synonymous in the latter half of the nineteenth century. Starting out during his internship at the *Hôpital du Midi* in the mid-1850s, Fournier's fervour for syphilology was to prove a lifelong fixation. In 1868, he became head of the venereology service at the *Hôpital Lourcine* which he served faithfully for eight years before becoming *Chef de Service* to the famous *Hôpital Saint-Louis*, then the undisputed mecca of nineteenth century French dermatovenereology (Fig. 2). As one of Paris' foremost venereologists, Fournier's was elected to occupy the second ever professorial chair of syphilology and dermatology in France shortly after it was erected by the Paris Faculty of Medicine on the last day of 1879 [9] – the first such chair was offered to Antoine Gailleton (1829-1904) at the Faculty of Medicine of Lyon in 1877 [10]. This dual professorship, although contested by a number of his colleagues, put Fournier in the position to learn and teach dermatology at Saint-Louis.

Many of Fournier's clinical contributions to dermatology were outlined by his famous student, Ferdinand Jean Darier (1856-1938), who detailed his master's works in a reverential obituary article written for him in mid-1915 [11]. From an English translation of Darier's article [12], we know that Fournier lectured on a number of purely dermatologic subjects (herpes, urticaria, and hydroa buccalis) and that he coined the term *diabétides* – a name that would come to encompass all the cutaneous manifestations of diabetes. His descriptions of vacciniform herpes in infants, drug eruptions due to antipyrine, fulminating gangrene of the genitalia, and recurrent buccal herpes in syphilitics were also offered as examples of his dermatologic writings. As others have suggested, Darier was apt to report that *the dermatological types which particularly attracted Fournier's attention are those which have some points of contact with syphilis*. This truism was many times amplified by Professor Louis Nékám (1868-1957), another student of Fournier's,

Figure 2: Alfred Fournier and colleagues in his famous dermatovenereology clinic at the *Hôpital Saint-Louis* in 1891. From the Collection *Bibliothèque interuniversitaire de Santé Médecine.*

who in September of 1935, faced members of the ninth international congress of dermatology in Budapest to make the now well-known satirical remark that Fournier classified skin diseases as *syphilitic, parasyphilitic, syphiloid and asyphilitic*! [13].

Dealing daily with syphilitic patients, Fournier was sure to document precisely, the many and varied syphilitic skin eruptions. This aspect of dermatology – pure dermatovenereology – was surely Fournier's forte, and Gaston Milian (1871-1945), Fournier's final chief of clinic, once recorded how *describing the cutaneous and mucous manifestations of syphilis is almost entirely his doing* [14]. In this regard, the syphilitic origin and precancerous nature of leukoplakia caught Fournier's close attention [15] and he became known for having classed this disease into a group he called the *parasyphilitic* affections [16] – the same group to which he famously designated tabes dorsalis and general paralysis. The cutaneous side effects of mercurial and iodide therapy likewise interested Fournier, and this author was not at all surprised to have found that Fournier had indeed written quite specifically on these topics. Along with mercurial stomatitis, specified dermatologic reactions outlined by Fournier in his lectures on mercurial therapy [17] include hydrargyria associated pruritus and the highly variable *desquamative polymorphous erythema* (amongst other idiosyncratic irritations). In discussing the evolving clinical features of the latter and the skin diseases with which it could be confounded – *scarlatina, measles, urticaria, eczema, erythema multiforme, erysipelas, commencing small-pox, exfoliating dermatitis*– Fournier demonstrates a degree of familiarity with even the asyphilitic dermatoses. His descriptions of skin reactions to iodide therapy were

just as nuanced [18], and the specific types he thus identified – namely *iodic acne*, *iodic purpura*, and other more severe *iodidides*– were very handsomely reported. In the latter class of more severe iodic skin reactions – the *'iodidides'* as they were then referred to – Fournier again demonstrates considerable dermatologic skill in classifying accurately the differing forms of the disease. He listed *bullous*, *furunculo-carbuncular*, *pustulo-crustaceous*, and *mycotic* subtypes, with the comment that the resemblance between *pustulo-crustaceous* iodic eruptions and certain tertiary syphilides could fool even the most careful clinician. In their syphilitic appearance, these iodic rashes sure were to fascinate Fournier and he was apt to tell his students how they *form one of the most interesting chapters in dermatology* [19]. Even outdated – the term *iododerma* has replaced much of the above given terminology – Fournier's work remains exemplary in its admirable degree of attention to clinical detail.

NON-CLINICAL CONTRIBUTIONS TO DERMATOLOGY

Although Fournier's contributions to clinical dermatology were limited in scope, he certainly played more important a managerial role in the development of the specialty in France. Such was his pivotal involvement in founding the *French Society of Dermatology and Syphilography*, which formed in June of 1889 and officially convened annual meetings in April of the following year [20]. From its very inception, Fournier contributed greatly to the society's work, and given that his presidential term – starting in the year of his retirement in 1902 – ran for a full seven years [21], we get some idea of the high esteem in which he was held by his fellow colleagues. The same society flourishes to this very day under the name of the *French Society of Dermatology and Sexually Transmitted Diseases*, and meets annually to discuss modern advances in both the fields of dermatology and venereology.

Apart from his involvement with France's premier dermatologic society, Fournier was also a central figure at the First World Congress of Dermatology, which was hosted by the *Hôpital Saint-Louis* in August of 1889 [22] (Fig. 3). Alfred Fournier was then at the peak of his productivity, and having penned numerous and authoritative texts on all aspects of syphilis, his works captured widespread attention from the international medical community. Of particular interest to the dermatologist is Fournier's friendly alliance with the English medical polymath Jonathan Hutchinson (1828-1913), who was, as far the study of syphilis is concerned, Fournier's equal in England. In 1886, Fournier honoured Hutchinson by coining the term *'Hutchinson's triad'* [23], and Hutchinson dedicated his textbook on syphilis

Figure 3: Alfred Fournier (seated centrally in front row) at the First World Congress of Dermatology in Paris, August 1889. The conference was held in Saint-Louis' historical hospital museum and hosted some 210 dermatologists from 29 different countries. Reproduced from *Dermatology in France* (p.415) with kind permission from the editors Dr. Daniel Wallach and Dr. Gérard Tilles.

to Fournier in the following year [24]. These gestures, alongside increasing international acclaim, made Fournier all the more famous back home in Paris, and many of Fournier's students took after their master in becoming notable figures in the history of dermatology themselves. Darier has already been referred to, but other widely regarded dermatologists to come from Fournier's clinic include Sabouraud, Wickham, Brocq, and Gaucher, to name but a few.

DERMATOVENEREOLOGIST OF THE CENTURY

Alfred Fournier has certainly been a favourite character amongst dermatovenereologists of the past century, and the above appellation is here given to him in recognition of his wholesome dedication to the craft. This is no place to detail the full gamut of Fournier's contributions to venereology, but it would be important to note that apart from his pioneering clinical advances in the field, it was the powerful human in Fournier that contributed so much to France's battle against syphilis in the pre-antibiotic era. A highly ethical man, Fournier invariably put French society first in his crusade against syphilis, and in dealing with the social aspects of the disease, he perched himself as a vocal and life-long defender of its most vulnerable and innocent victims (Fig. 4). On top of his many clinical breakthroughs and worldwide influence, it was this aspect of his that had him widely commemorated in May of 1932, when an international conference on syphilis in Paris was organized in honour of his one hundredth birthday. The centenary of his death has been less widely celebrated, but recognized it has been, not only in this article, but also in a historical video tribute uploaded to *YouTube* [25] and a privately published book length biographical reflection written in his memory [26]. To those interested, such contributions may serve as a palatable introduction to the life and legacy of one whose name will forever be cherished as a practitioner *par excellence* in the annals of dermatovenereology.

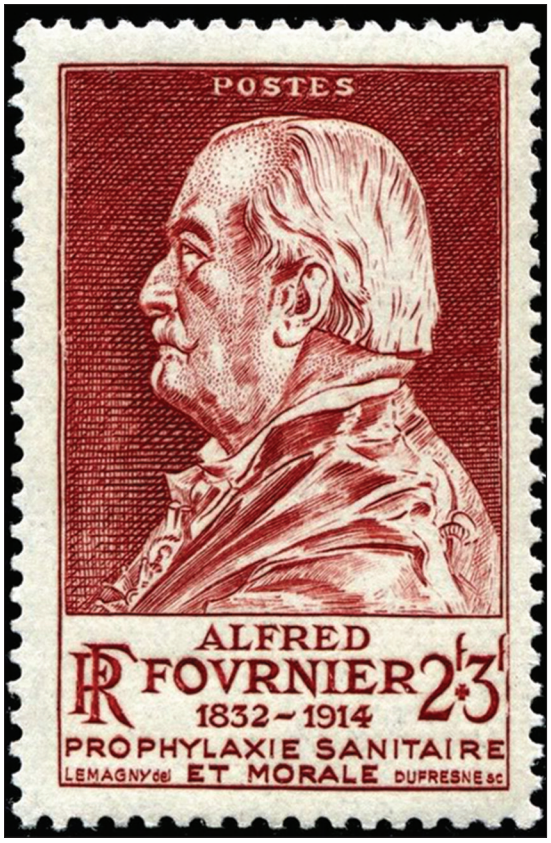


Figure 4: As a founder and inaugural president of the *The French Society for Sanitary and Moral Prophylaxis*, Fournier devoted himself to protecting French society from the ravages of syphilis. This commemorative postage stamp was issued by the society in February of 1946. From www.stampcommunity.org.

REFERENCES

1. Crissey JT, Parish LC, The Dermatology and Syphilology of the Nineteenth Century. New York: Praeger. 1981. Chapter 5: Lues I. p. 80.
2. Smith GL, Bunker CB, Dinneen MD. Fournier's gangrene. Br J Urol. 1998;81:347-55.
3. Lewis RT. Necrotizing soft-tissue infections. Infect Dis Clin North Am. 1992;6:693-703.
4. Bosshardt TL, Henderson VJ, Organ CH Jr. Necrotizing soft-tissue infections, in RG. Holzheimer, JA. Mannick (eds.) Surgical Treatment, Evidence-Based and Problem-Oriented, Zuckschwerdt Verlag GmbH: Munich.Presentation and Diagnosis section. 2001
5. Bunker CB, Neill SM. Chapter 71: The Genital, Perianal, and Umbilical Regions, in T. Burns, S. Breathnach, N. Cox, C. Griffiths (eds.) Rook's Textbook of Dermatology 8th Edition, Sydney: Wiley-Blackwell. 2010, p.71.31, Table 71.22.
6. Corbus BC, Harris FG. Erosive and gangrenous balanitis. The fourth venereal disease. JAMA. 1909;52:1474-7.
7. Fournier JA. Gangrène foudroyante de la verge. La Semaine Médicale, 1883 Dec; 3(56): 345-7. An English translation of this article was produced by Alexander S. Corman in 1988 and widely popularised in an article edited by his father Marvin Corman. See Dis Colon Rectum. 1988;31: 984-8.
8. Fournier JA. Etude clinique de la gangrène foudroyante de la verge. La Semaine Médicale. 1884;4:69-70.
9. Tilles G. The Hôpital Saint-Louis From 1607 Until 1945, in D. Wallach, G. Tilles (eds.) Dermatology in France. Editions Privat. Paris: Pierre Fabre Dermo-Cosmétique. 2002. p. 411. Towards official recognition of dermatology.
10. Chevallier J. Antoine Gailleton (1829-1904). une double vocation: Professeur de dermato-vénéréologie et maire de Lyon. Communication à la Société française d'Histoire de la Dermatologie, le 4 décembre 1998. Retrieved March 2015 from: http://sfhd.chez.com/ecrits/gaille.htm.
11. Darier FJ. Alfred Fournier 1832-1914. Ann Dermatol Syphil. 1915;5:513-28.
12. Darier FJ, Lane JE. Necrologie: Alfred Fournier 1832-1914. By J. Darier, Médecin de l'Hôpital Saint Louis, Translated with the author's permission, by John E. Lane, M.D., New Haven, Conn.

J Cut Dis Incl Syphil. 1918;36:482-93.
13. Nékám L. Un voyage d'étude dermatologique à la fin du XIXème siècle, in L. Nékám, Deliberationes Congressus Internationalis IX-I, Budapestini 13-21 Septembre 1935, Vol. IV. Budapesteni: Institutum Typographicum Patria. 1935. p. 185-209. Remark made on page 200.
14. Milian G. Le Professeur Alfred Fournier (1832-1914). Paris Médicale. 1916; 20: 133-137. Remark made on page 134, left column, 4th paragraph of new section.
15. Fournier JA. Des relations de la leucoplasie buccale avec la syphilis et le cancer. Tr. 13 Internat. Cong Med. 1900;9:496.
16. Fournier JA. Les Affections Parasyphilitiques. Paris: Rueff et Cie. 1894.
17. Fournier JA, Marshall CF. The Treatment of Syphilis; By Professor Alfred Fournier. English Translation.by C.F. Marshall, M.D., F.R.C.S. London: Rebman Limited. Chapter VII, Mercury, 1906, p. 51-56, 59-62.
18. Fournier JA, Marshall CF. Op Cit. Chapter XX, Iodide of Potassium. 1906, p. 184, 187, 189-191.
19. Ibid. p.189.
20. Civatte J. The French Society of Dermatology and Syphilology, in D. Wallach, G. Tilles (eds.) Dermatology in France. Editions Privat. Paris: Pierre Fabre Dermo-Cosmétique. 2002. p. 286-287.
21. Ibid. p. 299.
22. Tilles G. Op Cit. p. 415, The Hôpital Saint-Louis, seat of world dermatology congresses. 2002.
23. Fournier JA. La Syphilis Héréditaire Tardive. Paris: G. Masson. Section X: Triade d'Hutchinson, 1886. p.63.
24. Hutchinson J. Syphilis. London, Paris, New York, and Melbourne: Cassell & Company Limited. The formal dedication reads: To Alfred Fournier, as a small expression of friendship and high esteem this work is dedicated. 1887.
25. https://www.youtube.com/watch?v=YLg9SQvFqlI. Alternatively, search 'Jean Alfred Fournier' on YouTube to find the video.
26. Toodayan N. (December 2014) Jean Alfred Fournier (1832-1914): 'Benefactor of Humanity'. Clark and Mackay: Brisbane (ISBN 9780992467821). A much more detailed account of Fournier's life and works with a full list of references can be found in this work.

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

Historical Article

Dermatology Eponyms – sign –Lexicon (Q)

Piotr Brzeziński[1,3], Jana Zímová[2], Ewelina Cywinska[3], Anca Chiriac[4,5]

[1]Department of Dermatology, 6th Military Support Unit, Ustka, Poland, [2]Department of Dermatology, University Hospital Ostrava, 17. listopadu 1790, 708 52 Ostrava-Poruba, Czech Republic, [3]"Trychoderm" Clinic, ul. Kossaka 22, Slupsk, Poland, [4]Department of Dermatology, Nicolina Medical Center, Iasi, Romania, [5]Department of Dermato-Physiology, Apollonia University Iasi, Strada Muzicii nr 2, Iasi-700399, Romania

Corresponding author: Piotr Brzezinski, MD PhD, E-mail: brzezoo77@yahoo.com

ABSTRACT

Eponyms are used almost daily in the clinical practice of dermatology. And yet, information about the person behind the eponyms is difficult to find. Indeed, who is? What is this person's nationality? Is this person alive or dead? How can one find the paper in which this person first described the disease? Eponyms are used to describe not only disease, but also clinical signs, surgical procedures, staining techniques, pharmacological formulations, and even pieces of equipment. In this article we present the symptoms starting with (Q) and other. The symptoms and their synonyms, and those who have described this symptom or phenomenon.

Key words: Eponyms, Skin diseases, Sign, Phenomenon

Q SIGN

Endocarditis, pneumonia, fever, and liver involvement caused by the zoonotic Coxiella burnetii found in many mammals, including cattle, sheep, cats, dogs, rodents, birds, and ticks. Also called Q or Query Fever [1].

QUARTER EVIL SIGN

Symptomatic Anthrax [2]

This is found in certain cases of disease in cattle, having a rapid course and nearly always a fatal issue. The disease has several names, such as "Black-leg," "Quarter evil," and, in Germany, "Rauschbrand".

The bacilli are thickish rods, which are mostly single. They have lively motion by means of numerous cilia. The bacillus grows readily on ordinary media, but it is strictly anaerobic. It produces spores which are thicker than the bacillus and lie nearer one end than the other, so that a somewhat club-shaped form is produced. It produces gas in stab-cultures, and is decolorized when treated by Gram's method, in both these respects contrasting with bacillus anthracis.

QUECKENSTEDT SIGN

An indication of the existence of something; any objective evidence of a disease, ie, such evidence as is perceptible to the examiningphysician, as opposed to the subjective sensations (symptoms) of the patient [3,4].

QUEEN ANNE SIGN

In hypothyroidism, sparse eyebrows laterally (loss of outer eyebrows 2/3).

Apparently it was fashionable to shave the lateral third of the eyebrow during the region of Queen Anne (1701-1714) in Great Britain [5]. The association with Anne of Denmark is based on portraiture, although history does not suggest that she suffered an underactive thyroid. The eponym is disputed by some, though it has been suggested that Anne of France, Anne of Brittany, Anne of Austria, Anne Boleyn and Anne of Cleves may all be eliminated as candidates [5,6]. Known as sign of Hertoghe [7,8].

How to cite this article: Brzezinski P, Zímová J, Cywinska E, Chiriac A. Dermatology Eponyms – sign –Lexicon (Q). Our Dermatol Online. 2015;6(4):492-495.
Submission: 21.12.2014; **Acceptance:** 04.06.2015
DOI:10.7241/ourd.20161.137

QUEEN ANNE

Anne of Denmark (1574-1619) was Queen consort of Scotland, England, and Ireland as the wife of James VI and I.

Anne of Cleves (German: 1515 -1557) was Queen of England from 6 January 1540 to 9 July 1540 as the fourth wife of King Henry VIII.

Anne Boleyn (c. 1501-1536) was Queen of England from 1533 to 1536 as the second wife of King Henry VIII and Marquess of Pembroke in her own right.

Anne of Austria (1601-1666) was queen consort of France and Navarre, regent for her son, Louis XIV of France, and a Spanish and Portuguese Infanta by birth.

Anne of Brittany (1477-1514) was a French queen who reigned as Duchess of Brittany from 1488 until her death.

Anne of France (or Anne de Beaujeu) (1461-1522) was the eldest daughter of Louis XI of France and his second wife, Charlotte of Savoy [9,10].

QUINCKE'S SKIN SIGN, GIANT URTICARIA; URTICARIA OEDEMATOSA

Quincke's edema or Angioedema is the rapid swelling (edema) of the dermis, subcutaneous tissue, mucosa and submucosal tissues (Fig. 1a and b). It is very similar to urticaria, but urticaria, commonly known as hives, occurs in the upper dermis [11,12].

HEINRICH IRENAEUS QUINCKE

German physician, 1842-1922 (Fig. 2). He was educated in Berlin where he also completed his medical studies in 1864. After a 'grand tour' that took him to Paris, Vienna and London, he was trained in Berlin, first in surgery and later in internal medicine, under Von Frerichs (1819-1885). In 1878, he became a professor of internal medicine in Berne; from 1883 he held the chair of medicine in Kiel, which he would hold for the next 30 years.

In 1882, he published a synthesis of several observations of 'acute, circumscribed oedema of the skin'. Quincke accurately described the clinical features and distinguished the familial from the sporadic forms. He was correct in attributing the condition to increased vascular permeability, but he surmised the causal factors

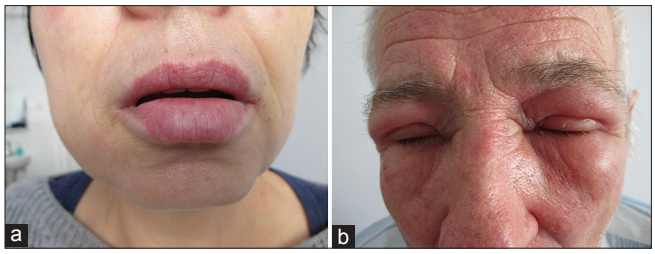


Figure 1: (a,b) Quincke's skin sign.

Figure 2: Heinrich Irenaeus Quincke.

were neurogenic rather than humoral, according to current insights (excess of bradykinin due to external factors or hereditary deficiency of C1-esterase inhibitor). Quincke not only contributed to several other clinical observations, but also pioneered the lumbar puncture, initially not for diagnostic purposes, but to relieve headache in hydrocephalic children [13,14].

QUINCKE'S SIGN

A blanching of the fingernails at each diastole of the heart. A sign seen in aortic insufficiency [11].

QUINQUAUD'S SCALP SIGN

A purulent folliculitis of the scalp, causing irregular bald patches (Figs 3a – f). Folliculitis decalvans is a

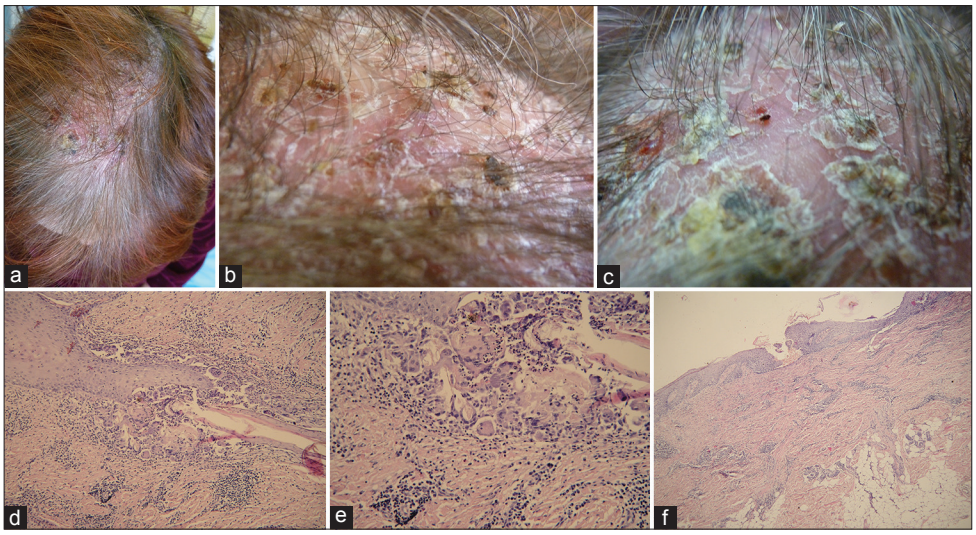


Figure 3: (a-c) Quinquaud's S\scalp sign, (d) The detail of the disintegrating hair follicle from picture one (FDCH 1), (e) Even bigger enlargement showing both elements: polymorphonuclears and giant multinucleated cells with fragments of keratin. Simultaneously, there can be seen plasmocytes in the surroundings of hair follicle, (f) The resulting status of scarring alopecia with disappearing hair follicle.

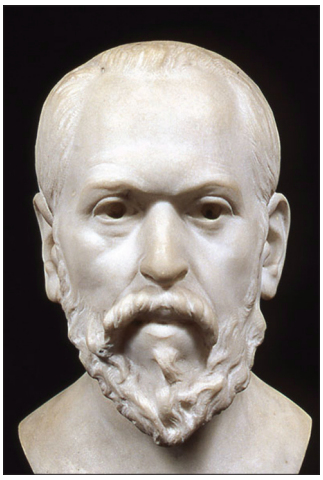


Figure 4: Charles Eugene Quinquaud (Bibliothèque de l'Académie nationale de médecine, 16 rue Bonaparte, 75006 Paris).

rare inflammatory scalp disorder. [15-18]. Folliculitis decalvans was first described by Quinquaud in the 1888. He reported one case of "folliculite épilante et destructive des regions velues" in 1888 [17]. He investigated three similar cases of pustular scarring alopecia and isolated bacteria from the hair follicle. Quinquaud transferred the bacteria to rats, mice, and rabbits but was not able to provoke a similar response in their fur. Brocq et al. in 1905 described Quinquaud's clinical findings under the designation "folliculitis decalvans" and distinguished it from other types of cicatricial alopecia [16].

CHARLES EUGENE QUINQUAUD

French physician (1841-1894) (Fig. 4). Charles Eugene Quinquaud entered medical school at Limoges in 1864 and in 1868 moved to Paris, where he obtained his doctorate in 1873. He was the last Interne des hôpitaux under Pierre Antoine Ernest Bazin (1807-1878) and it was Bazin who influenced him to study dermatology. He became médecin des hôpitaux in 1878, agrégé in 1883, and in 1886 chef de service at the Hôpital Saint-Louis, with Ernest Henri Besnier (1831-1909), Jean Alfred Fournier (1832-1915), and Jean Baptiste Emile Vidal (1825-1893) as colleagues. He was elected member of the Académie de Médecine in 1892.

Quinquaud contributed to many areas of medicine, being a skilled bacteriologist as well as a clinician, and with Nestor Gréhant (1838-1910) he developed a method for measuring blood volume using carbon monoxide (1882). He gave popular courses in internal medicine and pathology as well as dermatology and unselfishly supported Louis Brocq, enabling him to teach and see patients despite considerable opposition by some who were jealous of this young man's meteoric success and popularity.

In 1880, 1885, and 1887 Quinquaud won academic prizes for his works. He was editor of the journal La médecine scientifique [19].

REFERENCES

1. Muniain Ezcurra MA, Gálvez-Acebal J. Q fever and fever of unknown origin. Are the chronic forms of these conditions preventable? Rev Clin Esp. 2015;215:274-5.
2. Kunii O, Kita E, Shibuya K. [Epidemics and related cultural factors for Ebola hemorrhagic fever in Gabon]. Nihon Koshu Eisei Zasshi. 2001;48:853-9.

3. Deliyannakis E. Influence of the position of the head on the cerebrospinal fluid pressure. Variations of the Queckenstedt sign. Mil Med. 1971;136:370-2.
4. Ikebe S, Yokochi F, Wada T, Arakawa A, Mori H, Suda K, et al. [A 66-year-old man with backache and progressive difficulty of gait]. No To Shinkei. 1993;45:981-90.
5. Keynes M. Letter to the editor. J Med Biogr. 2009;17:62.
6. Lane Furdell E. Eponymous, anonymous: Queen Anne's sign and the misnaming of a symptom. J Med Biogr. 2007;15:97-101.
7. Brzezinski P, Pessoa L, Galvão V, Barja Lopez JM, Petrovitch Adaskevich U, Niamba PA, et al. Dermatology Eponyms – sign –Lexicon (H). Our Dermatol Online. 2013;4:130-43.
8. Al Aboud K. Medical Eponyms linked to hair. Our Dermatol Online. 2012;3:377-8.
9. Ross J. Queen Anne (1665-1714) and her health. J Med Biogr. 2015;23:54-9.
10. Regöly-Mérei G. [Paleopathological study of the remains of Béla the Third, King of Hungary, and his wife, Queen Anne]. Orv Hetil. 1968;109:423-7.
11. Giavina-Bianchi P, Aun MV, Motta AA, Kalil J, Castells M. Classification of angioedema by endotypes. Clin Exp Allergy. 2015;45:1142-3.
12. van Gijn J, Gijselhart JP. [Quincke and his oedema]. Ned Tijdschr Geneeskd. 2012;156:A5238.
13. Cozanitis DA. Heinrich Irenaeus Quincke (1842-1922): the Nobel Prize but for the problem of age. Presse Med. 2013;42:464-70.
14. Minagar A, Lowis GW. Dr Heinrich Irenaeus Quincke (1842-1922): clinical neurologist of Kiel. J Med Biogr. 2001;9:12-5.
15. Faye I, Marchand JP, N'Diaye B, Sarrat H. [Atypical form of Quinquaud's folliculitis decalvans]. Bull Soc Med Afr Noire Lang Fr. 1973;18:189-90.
16. Otberg N, Kang H, Alzolibani AA, Shapiro J. Folliculitis decalvans. Dermatol Ther. 2008;21:238-44.
17. Quinquaud E. Folliculite epilante et destructive des regions velues. Bull Mem Soc Hop Paris. 1888;5:395-8.
18. Zímová J, Pock L. Folliculitis decalvans capillitii. Dermatol Praxi. 2012;6:26-8.
19. http://www.whonamedit.com/doctor.cfm/1105.html

Source of Support: Nil, **Conflict of Interest:** None declared.

Our Dermatology Online

www.odermatol.com

4.2015 (28.September.2015)