E-ISSN 2651-5164

Türkderm

Türkderm - Türk Deri Hastalıkları ve Frengi Arşivi

2

DERİ VE ZÜHREVİ HASTALIKLAR DERNEĞİ 1930

www.turkderm.org.tr

Haziran/June 2025

CİLT-VOLUME 59

Türkderm

Türkderm - Türk Deri Hastalıkları ve Frengi Arşivi

Yayın Kurulu / Editorial Board

Yazışma Adresi / Address for Correspondence

Deri ve Zührevi Hastalıklar Derneği

Cerrahpaşa Mahallesi Kocamustafapaşa Caddesi No:106 D:1 Cerrahpaşa, Fatih, İstanbul, Türkiye
Telefon/Faks: +90 212 589 63 36 | turkderm@turkderm.org.tr

Dergide yayınlanan makalelerin, dergi standartlarına uygunluğunun kontrolü, dizimi, ingilizce makale ve özetlerin, kaynakların kontrolü, düzeltilmesi, derginin yayına hazır hale getirilmesi ve baskısı Galenos Yayınevi Tic. Ltd. Şti. tarafından gerçekleştirilmiştir.

Bu dergide kullanılan kağıt ISO 9706: 1994 standardına uygundur. (Requirements for Permanence) National Library of Medicine biyomedikal yayınlarda asitsiz kağıt (acid-free paper/ alkalin kağıt) kullanılmasını önermektedir.

The paper used to print this journal conforms to ISO 9706: 1994 standard (Requirements for Permanence). The National Library of Medicine suggests that biomedical publications be printed on acid-free paper (alkaline paper).

Yayınevi İletişim/Publisher Contact
Adres/Address: Molla Gürani Mah. Kaçamak Sk. No: 21/1 34093 İstanbul, Türkiye
Telefon/Phone: +90 (530) 177 30 97
E-posta/E-mail: info@galenos.com.tr/yayin@galenos.com.tr
Web: www.galenos.com.tr
Yayıncı Sertifika Numarası/Publisher Certificate Number: 14521

Basım Yeri/Printing at: Özgün Ofset Ticaret Ltd. Şti.
Yeşilce Mah. Aytekin Sk. No: 21 34418 4. Levent, İstanbul, Türkiye
Telefon/Phone: +90 (212) 280 00 09 **Sertifika No:** 42419

Yayın Tarihi/Publication Date: Temmuz 2025/July 2025

E-ISSN: 2651-5164

Üç ayda bir yayımlanan süreli yayındır. / International scientific journal published quarterly.

Türkderm

Türkderm - Türk Deri Hastalıkları ve Frengi Arşivi

Derginin "Yazarlara Bilgi" ve "Hakem Değerlendirmesi ve Etik" konularında bilgi almak için lütfen web sayfasına **(https://journal.turkderm.org.tr/tr/)** başvurun.

Yayın politikaları "Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals (ICMJE Recommendations)" (2016, http://www.icmje.org/) temel alınarak hazırlanmıştır. Türkderm Dergisi, **Web of Science-Emerging Sources Citation Index (ESCI), Hinari, GOALI, ARDI, OARE, AGORA, Excerpta Medica/EMBASE, EBSCO Database, Gale, CINAHL, Index Copernicus, Scopus, ProQuest, CABI, British Library, J-Gate, IdealOnline, Türk Medline-Ulusal Atıf İndeksi, Türkiye Atıf Dizini** ve **TÜBİTAK/ULAKBİM** tarafından indekslenmektedir.

Dergi, çevrimiçi olarak yayınlanmaktadır.

Sahip: Deri ve Zührevi Hastalıklar Derneği adına Ertuğrul Hasbi Aydemir

Sorumlu Yönetici: Fatih Göktay

Please refer to the journal's webpage **(https://journal.turkderm.org.tr/)** for "Instructions to Authors" and "Peer Review and Ethics".

The editorial policies are based on the "Recommendations for the Conduct, Reporting, Editing, and Publication of Scholarly Work in Medical Journals (ICMJE Recommendations)" by the International Committee of Medical Journal Editors (2017, archived at http://www.icmje.org/) rules. Turkderm-Turkish Archives of Dermatology and Venereology is indexed in **Web of Science-Emerging Sources Citation Index (ESCI), Hinari, GOALI, ARDI, OARE, AGORA, Excerpta Medica/EMBASE, EBSCO Database, Gale, CINAHL, Scopus, ProQuest, CABI, British Library, J-Gate, IdealOnline, Turkish Medline-National Citation Index, Turkiye Citation Index** and **TUBITAK/ULAKBIM**.

The journal is published online.

Owner: Ertuğrul Hasbi Aydemir on the behalf of the Society of Dermatology and Venereology

Responsible Manager: Fatih Göktay

Türkderm

Türkderm - Türk Deri Hastalıkları ve Frengi Arşivi

İçindekiler

Türkderm

Türkderm - Türk Deri Hastalıkları ve Frengi Arşivi

Contents

Original Investigation
Orijinal Araştırma

DOI: 10.4274/turkderm.galenos.2025.52333
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):39-44

Clinico-mycological assessment of pediatric dermatophytosis: experience of an eastern Indian tertiary hospital

Pediatrik dermatofitozun klinik-mikolojik değerlendirmesi: Doğu Hindistan üçüncü basamak hastanesinin deneyimi

Arya Bose*, Abheek Sil, Loknath Ghoshal*****

*Institute of Child Health, Department of Dermatology, Institute of Child Health, Kolkata, India
**PKG Medical College and Hospital, Department of Dermatology, West Bengal, India
***Malda Medical College, Department of Dermatology, West Bengal, India

Abstract

Background and Design: The ongoing rampage of dermatophytic infection continues to challenge dermatologists with its varied presentations, sparing no age group. Thus, this study was undertaken to assess the clinical, demographic, and microbiological aspects of childhood tinea infection.
Materials and Methods: A descriptive, cross-sectional study was conducted in 200 consecutive pediatric patients (under 14 years of age) diagnosed with dermatophytosis between January 2019 and March 2020. All patients underwent detailed history, thorough clinical examination, microbiological confirmation with potassium hydroxide mount and fungal culture.
Results: The majority of the study population (44.5%) belonged to the 3 ≤9 years age group with a mean age of 6.1 ± 3.3 years, with an overall male predominance. More than one-third of the patients (39%) belonged to the 'low' socioeconomic stratum and lived in rural areas. Similar affliction among family members was recorded in 83.5%. Chronic and recurrent dermatophytosis was diagnosed in 9% of patients. The most common site involved was the trunk. Clinically, the most commonly diagnosed condition was tinea corporis (92%). More than half of the patients (53.5%) reported usage of prior topical steroid-laced creams. The most common fungus was Trichophyton mentagrophytes.
Conclusion: A substantial pediatric population is afflicted by the ongoing scourge of superficial dermatophytosis. Results from our study corroborate with the studies done by Indian investigators on adult patients suffering from dermatophytosis. Multicentric collaboration with added molecular studies assessment is needed to address this growing concern in the pediatric age group.
Keywords: tinea, childhood, fungal, epidemiology, microbiology, epidemic

Öz

Amaç: Dermatofitik enfeksiyonun süregelen yaygınlığı, çeşitli klinik görünümleri ile, her yaş grubunu etkileyerek dermatologları zorlamaya devam etmektedir. Bu nedenle, bu çalışma çocukluk çağı tinea enfeksiyonunun klinik, demografik ve mikrobiyolojik yönlerini değerlendirmek amacıyla yapılmıştır.
Gereç ve Yöntem: Ocak 2019 ile Mart 2020 tarihleri arasında dermatofitoz tanısı almış 200 ardışık pediatrik hastada (14 yaş altı) tanımlayıcı, kesitsel bir çalışma yürütüldü. Tüm hastalara ayrıntılı öykü, kapsamlı klinik muayene, potasyum hidroksit ile direkt inceleme ve mantar kültürü ile mikrobiyolojik doğrulama yapıldı.
Bulgular: Çalışma popülasyonunun çoğunluğu (%44,5) 3 - < 9 yaş grubuna aitti ve ortalama yaş 6,1 +/- 3,3 yıldı ve genel olarak erkek egemenliği vardı. Hastaların üçte birinden fazlası (%39) 'düşük' sosyoekonomik tabakaya aitti ve kırsal alanlarda yaşıyordu. Hastaların %83,5'inde aile bireyleri arasında da benzer bir hastalık kaydedildi. Hastaların %9'unda kronik ve tekrarlayan dermatofitoz tanısı kondu. En sık tutulan bölge gövde olarak tespit edildi. Klinik olarak en sık teşhis edilen durum tinea korporis idi (%92). Hastaların yarısından fazlası (%53,5) daha önce topikal steroid içeren kremler kullandığını bildirdi. En sık rastlanan mantar türü Trichophyton mentagrophytes'ti.

Address for Correspondence/Yazışma Adresi: Abheek Sil MD, DNB, SCE, MNAMS, PKG Medical College and Hospital, Department of Dermatology, West Bengal, India
E-mail: abheek.sil@gmail.com **ORCID:** orcid.org/0000-0002-8043-2721
Received/Geliş Tarihi: 09.04.2024 **Accepted/Kabul Tarihi:** 24.03.2025 **Publication Date/Yayınlanma Tarihi:** 01.07.2025

Cite this article as/Atıf: Bose A, Sil A, Ghoshal L. Clinico-mycological assessment of pediatric dermatophytosis: experience of an Eastern Indian tertiary hospital. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):39-44

Sonuç: Geniş bir pediatrik popülasyon, süregelen yüzeysel dermatofitoz sorunundan etkilenmektedir. Çalışmamızdan elde edilen sonuçlar, Hintli araştırmacılar tarafından dermatofitozdan etkilenen yetişkin hastalar üzerinde yapılan çalışmalarla desteklenebilir. Pediatrik yaş grubunda giderek artan bu endişeyi gidermek için moleküler çalışmaların da eklendiği çok merkezli işbirliğine ihtiyaç vardır.
Anahtar Kelimeler: pediatri, tinea, mantar, epidemiyoloji, mikrobiyoloji, salgın

Introduction

Dermatophytosis is a fungal infection that invades the keratinized tissues of the host (skin, hair, and/or nails).[1] Once considered an easy-to-manage condition, dermatophytoses have over the past decade emerged as a nagging public health problem.[2] Unscientific usage of topical steroid and fixed-drug combination creams, emergence of Trichophyton mentagrophytes as a causative pathogen, and the growing emergence of antifungal resistance (particularly to terbinafine) have added fuel to this fire.[3,4] Furthermore, the chronicity, financial burden, visibility of the disease (in exposed body parts), social ostracism, and discriminatory attitude have negatively impacted the psychological well-being and quality of life in such patients.[5,6] The increasing prevalence of dermatophytoses among family members, including newborns and young children, presents a challenge not only for treating pediatricians but also trained dermatologists. Understanding the disease burden in a tertiary care centre (catering to a large catchment area) is crucial due to the associated high morbidity.

This study, emphasizing the demographic, clinical, and microbiological parameters of dermatophytoses in the pediatric population in an Eastern Indian tertiary care teaching hospital, was conducted to address this existing knowledge gap in the literature.

Materials and Methods

This was an institution-based, descriptive, cross-sectional study, conducted at a tertiary care teaching hospital in eastern India over one year (January 2019 to March 2020) after Institutional Ethics Committee clearance and in accordance with Good Clinical Practices and Helsinki's Declaration. The sample size was derived with the help of the OpenEpi software (OpenEpi, Atlanta, USA) after taking the reference frequency value from another study.[7] Consecutive 200 pediatric patients, aged less than 14 years, attending the dermatology OPD directly or referred from other departments, clinically diagnosed (by two dermatologists independently) with dermatophytosis, followed by microbiological confirmation, were recruited in the study (after informed and written consent from the guardian). All recruited patients underwent a thorough history (with special emphasis on socio-demographic profile and past treatment history). Clinical examination, routine biochemical investigations, and microbiological evaluation were done.

Microbiological assessment: In all cases, skin scrapings were collected from the edge of the lesion/ plucked hair/ nail clipping with a sterile scalpel after the affected areas (most representative and accessible site as determined by the treating dermatologist) were decontaminated with 70% alcohol. These specimens were taken on a clean, grease-free slide and mounted in 10-30% potassium hydroxide (KOH) to be examined under a light microscope for fungal elements after 5-10 minutes. All the samples were subjected to KOH preparation for a direct microscopic examination. Three drops of 10% KOH plus 40% dimethyl sulfoxide were placed over a clean, grease-free slide with skin scraping samples placed on that and covered with a coverslip. Slides were examined in light microscopy, and hyaline branching septate hyphae and arthrospores were identified. For fungal culture, the isolates were grown on 4% Saboraud's Dextrose agar with chloramphenicol and cycloheximide slant tubes and dermatophyte test media. Incubation was done aerobically at 37 °C (for isolation of T. verrucosum) and at 28 °C for other dermatophytes. Cultures were observed, every alternate day, to check for the appearance of any fungal growth or production of any pigment over the reverse side of the slant. Lactophenol cotton blue (LCB) staining was done of smears from culture-positive slants to detect the presence of macro and microconidia. SDA slants were inspected for up to 4 weeks for growth. Culture was identified on the basis of its macro and microscopic features with LCB staining and urease test. The final diagnosis was confirmed by the positivity of the direct smear and culture.

Statistical Analysis

Data were recorded in a clinical record form and entered into an MS Excel worksheet, and descriptive statistical analysis was done by SPSS for Windows (version 13.0, Chicago, IL). Digital photographs were taken by a digital image recorder on the first visit.

Results

Demography

Among 200 patients, the majority (31%) belonged to the 9 to <14 years age group, followed by the pre-school age group (23%), the toddler stage and the infant stage. There was a male preponderance (56%) with a male: female ratio of 1.27:1. Mean age was 6.1 ± 3.3 years (range 3 months to 13 years). Around two-thirds (65%) of the study population were school-going children. As per the modified Kuppuswamy scale (2020),[8] a fair number (66.5%) of patients were socioeconomically challenged (belonging to the "lower" and "lower middle" strata of society). A rural background was appreciable in most patients (Table 1).

Relevant history

Chronic and recurrent dermatophytosis was established in 9% of the total patients. Afflicted family members (both recurrent and recent past) were noted in 83.5% of total patients. Prior treatment was received by the majority (85%) of patients. More than half (53.5%) of patients reported using over-the-counter topical steroid-laced preparations. Systemic antifungal (often at adult dosage) was noted in 19 (9.5%) patients. The majority had been treated by a pediatrician (34%), followed by quacks (19.5%), and self-medication by guardian/ parents (18%).

Clinical findings

Disease duration ranged from 4 days to 18 months (mean: 111.93 ± 28.52 days). About one-third of patients (34.5%) had 5/or more lesions. We observed at least one lesion measuring >10 cm in 22.5%, with a mean size of 7.19 cm ± (range from 2 to 62 cm).

Table 1. Characteristics of study population

Parameters	Frequency	Percentage
Age group (years)		
0 ≤1	19	9.5
1 ≤3	31	15.5
3 ≤6	46	23.0
6 ≤9	43	21.5
9 ≤14	61	30.5
Sex		
Male	112	56
Female	88	44
Residence		
Semi-urban	96	48
Urban	73	36.5
Rural	31	15.5
Socioeconomic status		
Lower	78	39
Upper lower	55	27.5
Lower middle	48	24
Upper middle	18	9
Upper	1	0.5
Family members affected		
Yes	167	83.5
No	33	16.5
Past history of similar illness		
Yes	18	9
No	182	91
Treatment history		
Treatment naïve	30	15
OTC steroid-antifungal creams	107	53.5
Topical antifungal	61	30.5
Systemic antifungal	19	9.5
Others	2	1
Physician consultation		
None	30	15
Self-medication	36	18
Quacks	39	19.5
Pediatrician	68	34
Dermatologist	27	13.5
Comorbidities		
None	188	94
Nutritional deficiency	12	6
Iatrogenic Cushings	2	1
Ventricular septal defect	1	0.5
Duration (months)		
<1	35	17.5
1 ≤3	56	28
3 ≤6	83	41.5
6 ≤12	20	10
≥12	6	3
Number of lesions		
<5	131	65.5
≥5 – 10	62	31
>10	7	3.5
Size of largest lesion (cm)		
<10	155	77.5
≥10	45	22.5
Sites affected		
Face + scalp	31	15.5
Axilla	23	11.5
Torso	146	73
Upper limbs	35	17.5
Lower limbs	41	20.5
Groin and genitalia	62	31
Acral areas including nails	10	5
Clinical diagnosis		
Tinea corporis	184	92
Tinea faciei	28	14
Tinea capitis	16	8
Tinea cruris	62	31
Tinea manuum	10	5
Tinea unguium	2	1
Erythroderma	3	1.5
Fungal isolate (*n=184)		
Trichophyton mentagrophytes	146	79.3
Trichophyton rubrum	29	15.8
Epidermophyton	8	4.3
Microsporum	1	0.6

The most common site of involvement was the torso (73%), followed by groin and genitalia (31%), lower limbs (20.5%), and others. The mean body surface area involved was 9.57% (range 1-40%). The most common clinical presentation was tinea corporis (92%), followed by tinea cruris (31%), tinea faciei (14%), and tinea capitis (8%), amongst others. The pattern of tinea pseudoimbricata (ring-within-ring) was appreciated in 58% of patients (Figure 1-4).

Microbiological Findings

KOH mount staining showed long, branching septate hyphae in 184 cases (92%). On fungal culture, the most isolated was T. mentagrophytes (79.3%), followed by T. rubrum (15.8%), Epidermophyton (4.3%), and Microsporum (0.6%) (Figure 5).

Discussion

Over the past decade, India has been plagued by the menace of superficial dermatophytosis. This difficult-to-treat problem has emerged as a major public health problem, with increased prevalence of altered clinical forms like chronic, recurrent, recalcitrant, and erythrodermic varieties[9,10]. In our study, we documented 200 evaluable cases of pediatric dermatophytosis.

Male preponderance is seen among the children, as in adults. Similar to our findings, sex ratio has been documented to range from 1.27:1 to 1.8:1 in different studies[11-15]. Age group 9-13 years (31%) was found to be most affected by the condition in our study. Data on dermatophytosis involving pediatric cohorts suggests the age group between 11 and 16 years to be commonly affected (Table 2)[11-15]. Work conducted on chronic and recurrent dermatophytosis by Zacharia and Kunjukunju[16] and Sharma et al.[17] reported 18% and 33.3% of the study group to be students, respectively[11,12]. A study conducted by Kar et al.[18] in a neonatal care set-up reported tinea corporis in 10.2% and tinea faciei in 1.8% of admitted neonates. In recent studies, infants and toddlers constituted 8.9-11.48% of the pediatric study group[11,13].

Turkderm - Turk Arch Dermatol Venereol
2025;59(2):39-44

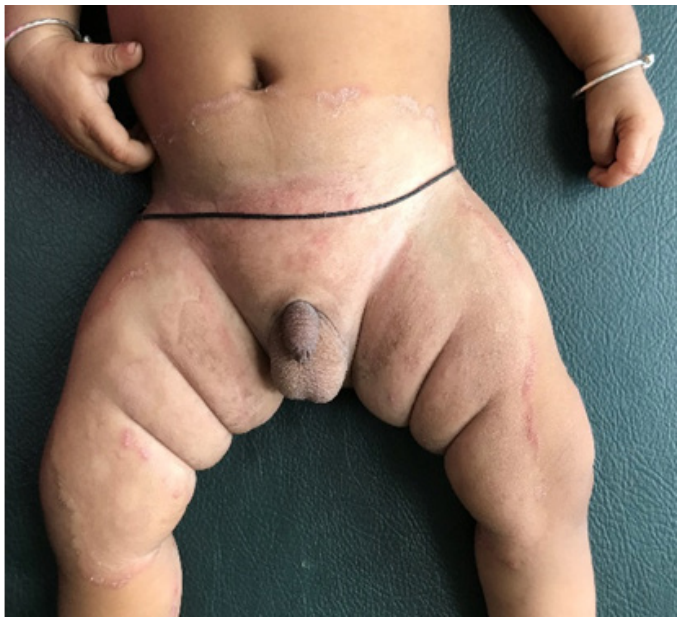

Figure 1. Tinea cruris in an infant

In congruence, our study found infants to comprise 9.5% of the study population. Dermatophytosis of the glabrous skin observed in neonates and infants speaks volumes of the gravity of the current epidemic-like scenario of dermatophytosis in India.

Various studies on dermatophytosis in children have documented the contact history with infected family members or close contacts to range from 62.2% to 91.9%, with mothers being the primary contact source in young children[11-15]. We observed an alarmingly high (86%) positive history of contact with affected family members. Sharing of fomites with the infected family members or close contacts, use of tight synthetic garments such as jeans and leggings, synthetic uniforms and track pants, poor personal hygiene, increased physical activity and overcrowding result in a perfect milieu for the multiplication of dermatophytes, especially among adolescents.

In recent years, the use of topical steroid creams, either alone or in combination with antifungal and antibacterial components, procured over the counter or as per prescription, in children has become a very common but alarming phenomenon. Around half (53.5%) of individuals mentioned the usage of steroid-laced preparations in our study. Studies have documented the abuse of TCS creams in children to be in the range of 51% to 94%[13-15].

The shift in the etiological agent from the more common T. rubrum to the virulent T. mentagrophytes complex observed among the adults has not spared children. Clinico-mycological studies on pediatric dermatophytosis conducted by Poojary et al.[15] and Ray et al.[11] revealed Trichophyton mentagrophytes complex to be the predominant etiological agent (73.7% and 73.18%, respectively). Trichophyton mentagrophytes complex was the most common isolate in our study, with a slightly higher prevalence (79.3%).

In the past, dermatophytosis in children was almost synonymous with tinea capitis. Similar to recent trends, we found multisite involvement with affection of the torso (73%) and genitalia (31%) as common findings. Clinically, tinea corporis was the most predominant (92%) diagnosis. Myriad morphologies of tinea corporis have been described, like polycyclic lesions, annular, eczematous/erythematous/psoriasiform

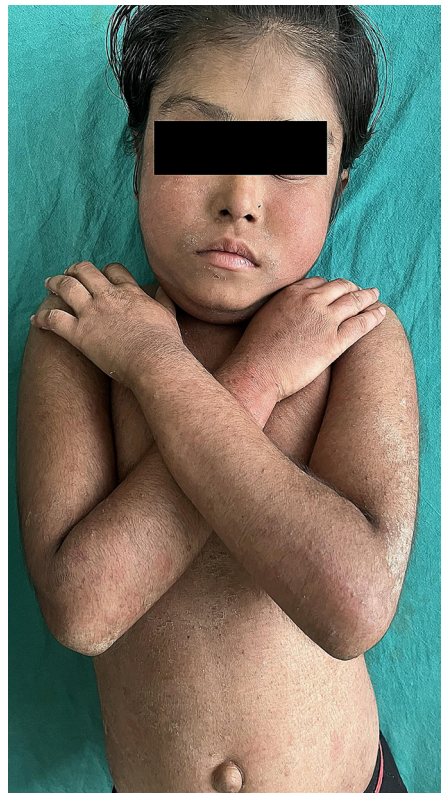

Figure 2. Cushingoid habitus in erythrodermic dermatophytosis

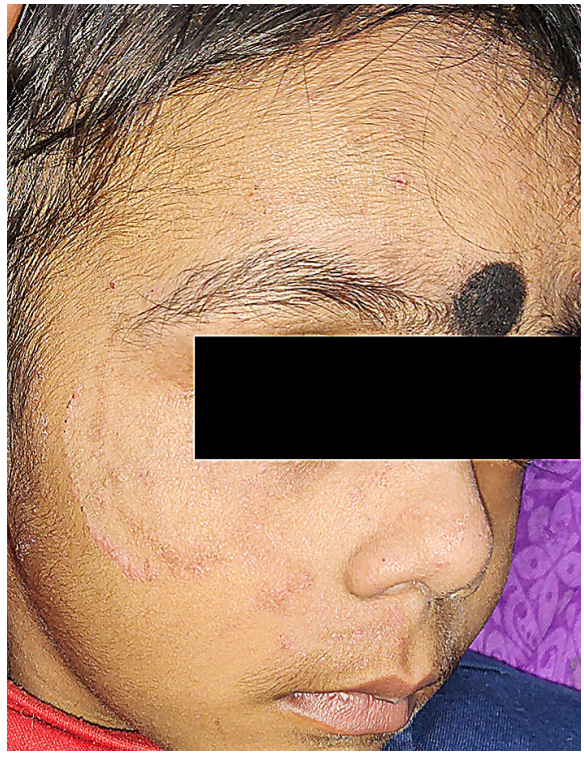

Figure 3. Steroid-modified tinea faciei showing multiple concentric rings (tinea pseudoimbricata)

Figure 4. Irritant contact dermatitis from home remedies superimposed on tinea corporis

Figure 5. Lactophenol cotton blue (LCB) mount showing septate hyphae with numerous spherical microconidia arranged in grape-like clusters, cigar-shaped macroconidia, and spiral hyphae

lesions, pustules, tinea pseudoimbricata, tinea incognito, and tinea recidivans[9,15]. While there has been an increase in the prevalence of tinea faciei, surprisingly, there is a decreasing trend of tinea capitis in the recent studies on pediatric dermatophytosis[19]. A pertinent observation in our study was the large proportion of children (58%) presenting with ring with ring pattern (pseudoimbricata) and some cases of erythrodermic variant (1%). Prior usage of topical steroids,

Table 2. Comparative analysis with other published Indian studies

Study/year (references)	Sample size	Study location	Study age group (yrs)	Age group commonly affected (%) /mean age (yrs)	Sex ratio	Positive family / contact history (%)	Treatment naïve (%)	Steroid usage (%)	Multisite involved (%)	Clinical diagnoses (%)	Fungal isolate (% positivity)
Dash et al.[12] 2017	198	Orissa	2-15	11-15 (51.5%) / NM	1.13:1	83.8	NM	-	16.2	Tinea cruris (50), Tinea corporis (47.47)	NS
Mishra et al.[13] 2018	235	Uttar Pradesh	<18	>6 – 9 (31.5%) / 7.7 ± 4.74	1.8:1	91.9	NM	94	27.2	Tinea corporis (45.5), Tinea cruris (29.4), Tinea faciei (24.3)	T. mentagrophytes (49)
Gandhi et al.[14] 2019	100	Karnataka	≤18	10-14 (56%) / NM	1.27:1	83%	32	51	NS	Tinea corporis (45), Tinea cruris (28), Tinea capitis (11)	NS
Poojary et al.[15] 2021	67	Maharashtra	<14	NS / 6.2 ± 4.3	1.68:1	76.1	11.9	85	52.2	Tinea corporis (73.1), Tinea cruris (55.2), Tinea faciei (17.9)	T. mentagrophytes (73.7)
Ray et al.[11] 2022	183	Orissa	<16	11-16 (47.5) / 9.5	1.22:1	62.2	NM	NS	55.2	Tinea corporis > Tinea cruris > Tinea faciei	T. mentagrophytes / interdigitale (73.18)
Our study	200	West Bengal	<14	3-6 years (44.5%)	1.27:1	83.5	15	53.5	64	Tinea corporis (92), Tinea cruris (31), Tinea faciei (14)	T. mentagrophytes (79.3)

Ref: Reference number; NM: Not mentioned; NS: Not studied, yrs: Years

unfortunately, was the attributable factor (Cushingoid features were observed in 2 patients).

In recent years, there has been an increasing spate of antifungal-resistant dermatophyte infections across the globe[20]. Although this resistance was initially noted in India, similar observations have now been reported across many parts of Europe, Iran, Japan, China, and more recently in the United States[21,22]. The predominant causative dermatophyte for these infections has been reported to be T. mentagrophytes genotype VIII, recently designated T. indotineae[22]. This fungal pathogen is responsible for chronic, recurrent, recalcitrant and widespread superficial infections. Resistance to terbinafine is frequently encountered and is related to point mutations in the gene encoding the squalene epoxidase[23].

Study Limitations

An important limitation in our study was our inability to assess the antifungal sensitivity pattern of isolates and carry out molecular studies due to resource constraints. Our hospital-based study carried out in a densely populated city in eastern part of eastern India may not be representative of the diverse clinico-mycological shift in other geographical regions.

Conclusion

Our study highlights the evolving trend in demographic profile and clinical features of childhood dermatophytosis in a part of Eastern India. The presence of a positive family history or close contact with an affected family member acts as an important source of pediatric affliction. Strict measures should be taken to discourage the use of over-the-counter topical medications. Education of the masses regarding seeking proper medical care by qualified personnel and maintaining hygiene in daily life is crucial. Further studies should be undertaken, particularly to explore whether there is any correlation between changing clinical patterns of presentation and species of fungal infection and rising antifungal unresponsiveness.

Ethics

Ethics Committee Approval: This was an institution-based, descriptive, cross-sectional study, conducted at a tertiary care teaching hospital in Eastern India over one year (January 2019 to March 2020) after Institutional Ethics Committee clearance and in accordance with Good Clinical Practices and Helsinki's Declaration.

Informed Consent: Written informed consent was obtained from all subjects included in this study including the use of their personal information and photographs to be printed for publication in a journal.

Footnotes

Authorship Contributions

Surgical and Medical Practices: A.B., A.S., L.G., Concept: A.B., A.S., L.G, Design: A.B., A.S., L.G.., Data Collection or Processing: A.B., A.S., L.G.., Analysis or Interpretation: A.B., A.S., L.G., Literature Search: A.B., A.S., L.G., Writing: A.B., A.S., L.G.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Verma SB, Panda S, Nenoff P, et al.: The unprecedented epidemic-like scenario of dermatophytosis in India: I. Epidemiology, risk factors and clinical features. Indian J Dermatol Venereol Leprol. 2021;87:154-75.
2. Panda S, Verma S: The menace of dermatophytosis in India: The evidence that we need. Indian J Dermatol Venereol Leprol. 2017;83:281-4.
3. Kumar S, Goyal A, Gupta YK: Abuse of topical corticosteroids in India: Concerns and the way forward. J Pharmacol Pharmacother. 2016;7:1-5.
4. Kanafani ZA, Perfect JR: Antimicrobial resistance: resistance to antifungal agents: mechanisms and clinical impact. Clin Infect Dis. 2008;46:120-8.
5. Das A, Fatima F, Sil A, Podder I, Jafferany M: Therapeutic management of psychological morbidity and impaired quality of life in patients with persistent dermatophytoses. Dermatol Ther. 2020;33:e14124.
6. Das A, Sil A, Fatima F, Podder I, Jafferany M: Impact of chronic and recurrent dermatophytosis on quality of life and psychologic morbidity-a cross-sectional study. J Cosmet Dermatol. 2022;21:3586-92.
7. Nagarajan K, Thokchom NS, Ibochouba K, Verma K, Bishurul Hafi NA: Pattern of pediatric dermatoses in Northeast India Indian J Paediatr Dermatol. 2017;18:286–91.
8. Ananthan VA: Modified Kuppuswamy scale for socioeconomic status of the Indian family- Update based on New CPI (IW) series from September 2020. J Family Med Prim Care. 2021;10:2048-9.
9. Dogra S, Narang T: Emerging atypical and unusual presentations of dermatophytosis in India. Clin Dermatol Rev. 2017;1:12–8.
10. Das A, Sil A, Jaiswal S, et al.: Erythrodermic dermatophytosis: an alarming consequence of steroid abuse and misuse. A multicentre prospective study from India. Clin Exp Dermatol. 2022;47:1735-8.
11. Ray A, Singh BS, Kar BR: Clinicomycological profile of pediatric dermatophytoses: an observational study. Indian Dermatol Online J. 2022;13:361-5.
12. Dash M, Panda M, Patro N, Mohapatra M: Sociodemographic profile and pattern of superficial dermatophytic infections among pediatric population in a tertiary care teaching hospital in Odisha. Indian J Paediatr Dermatol. 2017;18:191-5.
13. Mishra N, Rastogi MK, Gahalaut P, Yadav S, Srivastava N, Aggarwal A:. Clinicomycological study of dermatophytoses in children presenting at a tertiary care center. Indian J Paediatr Dermatol. 2018;19:326–30.
14. Gandhi S, Patil S, Patil S, Badad A:. Clinicoepidemiological study of dermatophyte infections in pediatric age group at a tertiary hospital in Karnataka. Indian J Paediatr Dermatol. 2019;20:52–6.
15. Poojary S, Jaiswal S, Bhalala KB, et al.: A cross sectional observational study of pediatric dermatophytosis: Changing clinico mycological patterns in Western India. Indian J Paediatr Dermatol. 2021;22:236–40.
16. Zacharia M, Kunjukunju BP: Clinical profile of patients with chronic dermatophytosis-A descriptive study from a tertiary care centre in Kerala. J Evid Based Med Healthc. 2017;4:2863–6.
17. Sharma R, Adhikari L, Sharma RL: Recurrent dermatophytosis:A rising problem in Sikkim, a Himalayan state of India. Indian J Pathol Microbiol. 2017;60:541–5.
18. Kar C, Sardar SK: Pattern of pathological cutaneous lesions of neonates in neonatal care unit of a peripheral tertiary institution in West Bengal. Indian J Paediatr Dermatol. 2019;21:22–30.
19. Rengasamy M: The changing scenario of dermatophytosis in children. Indian Dermatol Online J. 2022;13:305-9.
20. Kruithoff C, Gamal A, McCormick TS, Ghannoum MA: Dermatophyte infections worldwide: increase in incidence and associated antifungal resistance. Life (Basel). 2023;14:1.
21. Hiruma J, Noguchi H, Hase M, et al.: Epidemiological study of terbinafine-resistant dermatophytes isolated from Japanese patients. J Dermatol. 2021;48:564-7.
22. Chen E, Ghannoum M, Elewski BE: Treatment-resistant tinea corporis, a potential public health issue. Br J Dermatol. 2021;184:164-5.
23. Das A, Sil A, Verma SB, Kumar S: Tinea pseudoimbricata: observations from a clinicoepidemiological and mycological study from eastern India. Clin Exp Dermatol. 2022;47:147-9.

Original Investigation
Orijinal Araştırma

DOI: 10.4274/turkderm.galenos.2025.23281
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):45-53

Assessment of YouTube videos about nail health and conditions in Turkish

Tırnak sağlığı ve hastalıkları ile ilgili Türkçe YouTube içeriklerinin değerlendirilmesi

Ozan Erdem, Vefa Aslı Erdemir, Elif Ekin Alpdoğan, Süveyda Teko, Ahmet Sait Şahin, Peyruze Gül Daye, Mehmet Salih Gürel

İstanbul Medeniyet University Faculty of Medicine, Department of Dermatology and Venereology, İstanbul, Türkiye

Abstract

Background and Design: As YouTube becomes more popular as a source of health information, concerns about the reliability of its content are also increasing. While many studies have evaluated health-related content on YouTube, nail-related videos remain under-reviewed. This study aims to assess the engagement and quality of Turkish-language YouTube videos on nail health and disorders, focusing on their subject, content, creators, and sources of information.

Materials and Methods: A cross-sectional content analysis was conducted on 500 Turkish YouTube videos, collected using relevant keywords. Video data, including upload dates, duration, views, likes, and comments, were retrieved via a Python script using the YouTube Data API. Videos were categorized by uploader and person providing information in the video, and content quality was assessed using the Global Quality Scale (GQS). Descriptive statistics and non-parametric tests were used for data analysis.

Results: The most common video topics were ingrown toenail (39.0%), onychomycosis (31.8%), and nail health and care (9.4%), with the latter receiving the highest engagement in terms of views, likes, and comments. Although healthcare providers were the primary sources of information in 67% of the videos, independent non-healthcare content creators and patients attracted the most interaction. Healthcare professionals, particularly dermatologists, provided higher quality information in the videos than non-healthcare creators, yet these videos received less engagement. Videos containing herbal therapies and alternative medicine garnered the most attention but also had the lowest GQS scores.

Conclusion: Turkish-language YouTube content on nail health and disorders is primarily dominated by non-healthcare creators, who attract higher engagement despite lower content quality. A contrast exists between audience interactions and video quality. Patients seeking health information on YouTube should exercise caution to avoid being misinformed. Healthcare professionals should enhance their online presence by creating accurate and engaging content to ensure patients can access reliable information.

Keywords: Dermatology, nail diseases, nail disorders, nail health, social media, YouTube

Öz

Amaç: YouTube, sağlık bilgisi kaynağı olarak daha popüler hale geldikçe, içeriklerin güvenilirliği konusunda endişeler de artmaktadır. Birçok çalışma YouTube'daki sağlıkla ilgili içerikleri değerlendirmiş olsa da tırnak ile ilişkili videolar yeterince incelenmemiştir. Bu çalışma, tırnak sağlığı ve hastalıklarıyla ilgili Türkçe YouTube videolarının etkileşimini ve kalitesini, konuları, içerikleri, içerik oluşturucuları ve bilgi kaynaklarına odaklanarak değerlendirmeyi amaçlamaktadır.

Gereç ve Yöntem: Çalışma kapsamında, ilgili anahtar kelimeler kullanılarak 500 Türkçe YouTube videosu üzerinde kesitsel bir içerik analizi yapılmıştır. Video verileri, yükleme tarihleri, süreleri, izlenme sayıları, beğeniler ve yorumlar dahil olmak üzere YouTube Data API kullanılarak bir Python betiği aracılığıyla toplanmıştır. Videolar, yükleyen kişi ve videoda bilgiyi sağlayan kişiye göre kategorize edilmiş ve içerik kalitesi Global Kalite Skalası (GQS) kullanılarak değerlendirilmiştir. Veri analizi için tanımlayıcı istatistikler ve parametrik olmayan testler kullanılmıştır.

Address for Correspondence/Yazışma Adresi: Ozan Erdem, Asst. Prof., İstanbul Medeniyet University Faculty of Medicine, Department of Dermatology and Venereology, İstanbul, Türkiye
E-mail: erdemozan@hotmail.com **ORCID:** orcid.org/0000-0002-6012-0528

Received/Geliş Tarihi: 11.09.2024 **Accepted/Kabul Tarihi:** 09.03.2025 **Publication Date/Yayınlanma Tarihi:** 01.07.2025

Cite this article as/Atıf: Ozan Erdem O, Erdemir VA, Alpdoğan EE. Assessment of YouTube videos about nail health and conditions in Turkish. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):45-53

Bulgular: En sık video konuları tırnak batması (%39,0), onikomikoz (%31,8) ve tırnak sağlığı ve bakımı (%9,4) olup, tırnak sağlığı ve bakımı konusu en yüksek etkileşimi (izlenme, beğeni ve yorum) almıştır. Videoların %67'sinde sağlık profesyonelleri ana bilgi kaynağı olmasına rağmen, bağımsız sağlık dışı içerik üreticileri ve hastalar en fazla etkileşimi çekmiştir. Sağlık profesyonelleri, özellikle dermatologlar, sağlık dışı içerik üreticilerine kıyasla daha yüksek kalitede bilgi sağlamış, ancak bu videolar daha az etkileşim almıştır. Bitkisel tedaviler ve alternatif tıpla ilgili videolar en çok ilgiyi görmüş, ancak en düşük GQS puanlarına sahip olmuştur.
Sonuç: Tırnak sağlığı ve hastalıkları ile ilgili Türkçe YouTube içeriği büyük ölçüde sağlık dışı içerik üreticileri tarafından domine edilmekte olup, bu kişiler daha düşük içerik kalitesine rağmen daha yüksek etkileşim çekmektedir. İzleyici etkileşimleri ve video kalitesi arasında belirgin bir fark bulunmaktadır. YouTube'da sağlık bilgisi arayan hastalar, yanlış yönlendirilmemek için dikkatli olmalıdır. Sağlık profesyonelleri, hastaların güvenilir bilgilere erişimini sağlamak amacıyla, doğru ve ilgi çekici içerikler oluşturarak çevrimiçi varlıklarını güçlendirmelidir.
Anahtar Kelimeler: Dermatoloji, tırnak hastalıkları, tırnak bozuklukları, tırnak sağlığı, sosyal medya, YouTube

Introduction

In the past year, nearly 6 out of 10 individuals have turned to the internet for health or medical information, reflecting the growing reliance on online platforms for healthcare guidance[1]. YouTube, as one of the most widely used video-sharing platforms, plays a prominent role in this trend by offering easily accessible and cost-free advice on a variety of health-related topics, including skin conditions. However, despite its accessibility, the accuracy and quality of health information on YouTube can vary significantly, often depending on the source of the content[2,3].

Given concerns about the credibility of the information patients access, numerous studies have been conducted to evaluate the reliability of YouTube videos. Among these, studies related to skin disorders have primarily focused on English-language content, examining conditions such as acne[4,5], atopic dermatitis[6,7], psoriasis[8,9], hidradenitis suppurativa[10], and alopecia areata[11]. However, only a limited number of studies have centered on Turkish-language YouTube content, specifically for acne[12] and psoriasis treatments[13]. Additionally, research on nail disorders is sparse, addressing only onychomycosis treatment[14] and nail biopsy procedures,[15] leaving many other nail conditions largely unexamined.

The primary aim of this study is to assess the quality and reliability of Turkish-language YouTube videos on nail health and disorders, focusing on their subjects, content, creators, and sources of information. As the use of online platforms for health information continues to grow, healthcare professionals need to evaluate the reliability of popular platforms like YouTube. This evaluation will help ensure patients access accurate and trustworthy information, supporting better health decisions and outcomes.

Materials and Methods

A cross-sectional content analysis study evaluated Turkish YouTube videos on nail health and disorders. The videos to be included in the study were obtained through a search on the YouTube platform on August 15, 2024, using the Turkish equivalents of the following predetermined keywords: "nail," "nail diseases," "nail disorders," "nail health," "nail fungus," "ingrown nail," "nail dystrophy," "nail tumor," and "nail melanoma." Videos related to acrylic or prosthetic nails, non-therapeutic nail cosmetics, and non-Turkish videos were excluded from the study. Ultimately, 500 relevant videos were included for evaluation (Figure 1). For each video, data were collected on the upload date, time since upload (in days and months), video length (in minutes and seconds), and the number of views, likes, and comments. A Python script utilizing the YouTube Data API was used to initially search and retrieve the exact data from the video URLs to ensure accuracy.

The individual or organization responsible for producing and uploading the video to the YouTube platform was recorded. Various categories were established accordingly, including "physicians," "non-physician

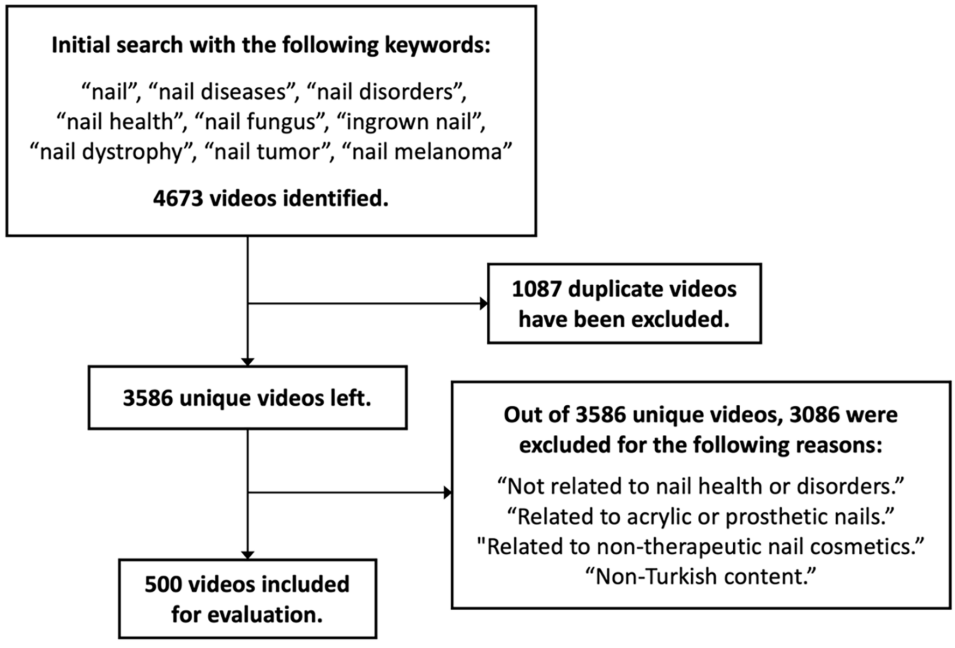

Figure 1. Flowchart of the search process for Turkish YouTube videos related to nail health and disorders

healthcare providers," "private healthcare institutions," "academic institutions," "online healthcare platforms," "television programs or national news agencies," "patients," and "independent non-healthcare content creators." In addition to documenting the uploader, the source of information presented in the video was also recorded, noting whether the individual providing the information in the video was a healthcare professional and, if so, their area of specialty.

The general quality of the videos was assessed for each video by two authors using the Global Quality Scale (GQS), originally developed by Bernard et al.[16] to evaluate medical website quality and also commonly applied to assess the quality of YouTube video content. The GQS is a five-point scale that examines video quality, flow, and usefulness. A score of 1 represents poor quality, indicating that the video is unlikely helpful for patients. A score of 2 reflects poor quality with some helpful information, though its utility for patients remains limited. A score of 3 indicates suboptimal flow, with some information covered but key topics missing, making the video somewhat useful for patients. A score of 4 represents good quality and flow, with the most important issues addressed, providing significant utility for patients. Finally, a score of 5 indicates excellent quality and flow, with comprehensive coverage of relevant topics, making the video highly useful for patients. When the two authors disagreed on a video's GQS score, they reached a consensus to determine the final score.

Statistical Analysis

All statistical analyses were performed using Statistical Package for the Social Sciences (SPSS) version 23.0 for Mac (SPSS Inc., Chicago, IL, USA). Descriptive values are presented as medians for non-normally distributed continuous variables and frequencies and percentages for categorical variables. Comparisons between two groups were performed using the Mann-Whitney U test for non-normally distributed data, while comparisons involving more than two groups were conducted using the Kruskal-Wallis H test. A p-value of less than 0.05 was considered statistically significant. Graphs were created using ChatGPT, a language model developed by OpenAI (OpenAI, 2024). The confidentiality of the individuals and organizations responsible for producing the videos was strictly maintained. Ethics committee approval was not required, as the study did not involve human or animal participants, and the videos were publicly accessible.

Results

The dataset spans videos uploaded to the YouTube platform between May 6, 2012 (approximately 12 years and 4 months ago) and August 13, 2024 (2 days ago), reflecting a broad range of content over more than a decade. The shortest video included is 23 seconds long, while the longest is 86 minutes and 46 seconds. The median video duration is 3 minutes and 14 seconds. The most frequent uploaders were independent non-healthcare content creators (27.4%), followed closely by physicians (27%) (Figure 2). Videos from private healthcare institutions (13.4%) and national media (11.6%) also contributed significantly. Healthcare providers, including both physicians and non-physician professionals, were the prominent persons who provided information in the majority of the videos (335/500, 67%), with physicians dominating this group (231/335, 69%) (Figure 3). Among physician-presented videos, dermatologists were the most prominent (33.8%), followed by specialists in orthopedics (26.8%), general surgery (19%), and internal medicine (6.1%) (Table 1). Notably, podiatry led the non-physician healthcare provider category (83.7%).

The most frequently covered subjects in the videos were ingrown toenails (39.0%), onychomycosis (31.8%), and nail health and care (9.4%) (Figure 4). Less common topics included nail tumors (2.0%), nail melanoma (1.4%), paronychia (1.2%) and nail psoriasis (0.4%). In terms of video content, general information dominated (34.2%), followed by surgical treatment (16.2%) and herbal therapies or alternative medicine (14.0%). The least common categories included medical treatment (4.4%) and laser treatment (1.8%) (Figure 4).

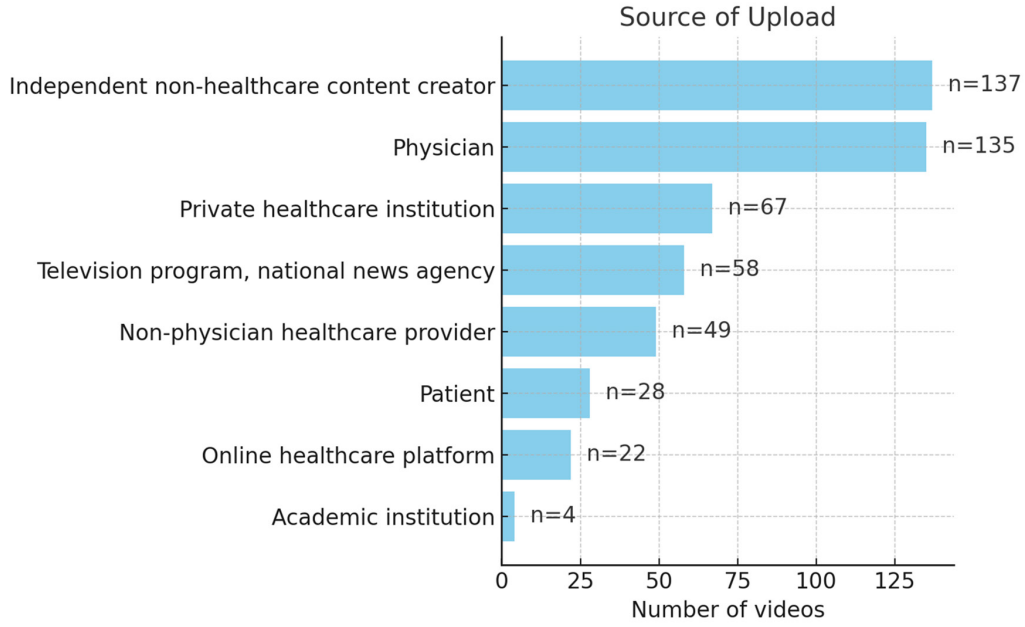

Figure 2. Distribution of YouTube videos by source of upload

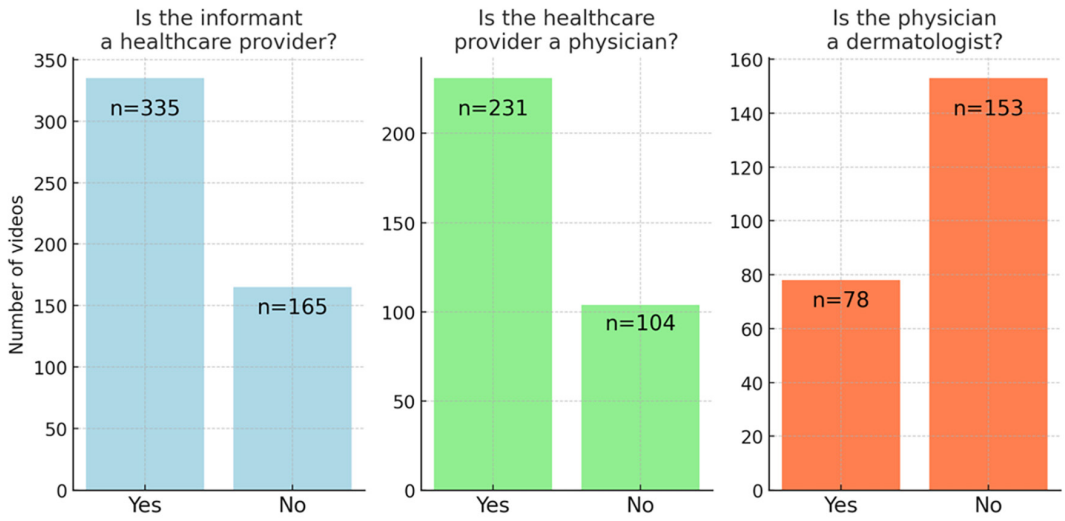

Figure 3. Classification of YouTube videos based on the expertise of the individual providing the information in the video

Table 1. Distribution of healthcare providers' areas of specialty among examined YouTube videos

Healthcare providers' area of specialty	Number of videos	
	%	n
Physician	231	100
Dermatology	78	33.8
Orthopedics	62	26.8
General surgery	44	19
Internal medicine	14	6.1
General practitioner	8	3.5
Psychiatry	8	3.5
Pediatrics	6	2.6
Cardiovascular surgery	5	2.2
Plastic surgery	4	1.7
Pediatric surgery	2	0.9
Non-physician healthcare provider	104	100
Podiatry	87	83.7
Pharmacy	5	4.8
Nursing	4	3.8
Aesthetics	4	3.8
Psychology	3	2.9
Nutrition	2	1.9

Video engagements were thoroughly examined based on the source of upload, video subject, and content. Independent non-healthcare content creators and patients generated the highest median views, likes, and comments, with independent creators receiving 51,051 views, 545 likes, and 31 comments and patients gathering 34,224 views, 173.5 likes, and 52.5 comments (Figure 5). In contrast, videos from academic institutions had the lowest engagement, with a median of only 1,396 views, 4.5 likes, and zero comments. The subject receiving the most interaction was nail health and care, with a median of 35,740 views, 357 likes, and 79 comments. Other topics like paronychia (17,684 views, 90.5 likes, 20 comments), onychomycosis (16,336 views, 90 likes, four comments), and ingrown toenail (14,131 views, 55 likes, nine comments) also attracted significant attention. In comparison, nail melanoma and nail tumors saw the least interaction (4,812 views, 18 likes, zero comments, and 4,030 views, 19 likes, and zero comments, respectively). Regarding content, herbal therapies, and alternative medicine stood out with the highest medians (98,740 views, 856 likes, and 38 comments).

Comparison between different individuals providing information in the videos revealed that informants other than healthcare providers had significantly higher interaction in terms of views, likes, and comments compared to healthcare professionals ($p<0.001$) (Table 2). Among healthcare professionals, physicians had more views and likes than non-physician healthcare providers ($p=0.015$ for views, $p=0.017$ for likes), although the number of comments was similar ($p=0.369$). Videos featuring dermatologists had fewer views, likes, and comments than those from other physician specialties, with all differences showing strong statistical significance ($p<0.001$).

The quality of videos was analyzed based on the upload source, video subject, and content. Videos uploaded by patients and independent non-healthcare content creators had the lowest GQS scores, with medians of 1.0 and 2.0, reflecting the poorest quality (Figure 6). In contrast, videos from academic institutions had the highest quality, with a median GQS of 5.0. For video subjects, nail health and care and onychomycosis had the lowest scores (medians of 2.0). In contrast, nail tumors and melanoma were rated the highest, with medians of 4.5 and 5.0. Regarding content, experience sharing and herbal therapies were rated the lowest (median of 1.0), while general information and laser treatment scored the highest, with a median of 4.0.

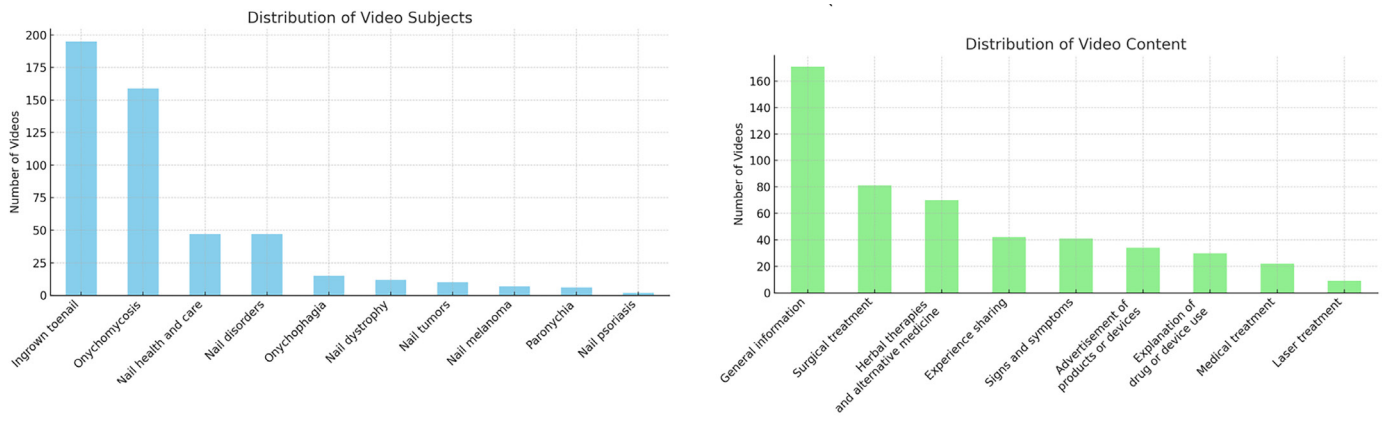

Figure 4. Distribution of YouTube videos by subject and content

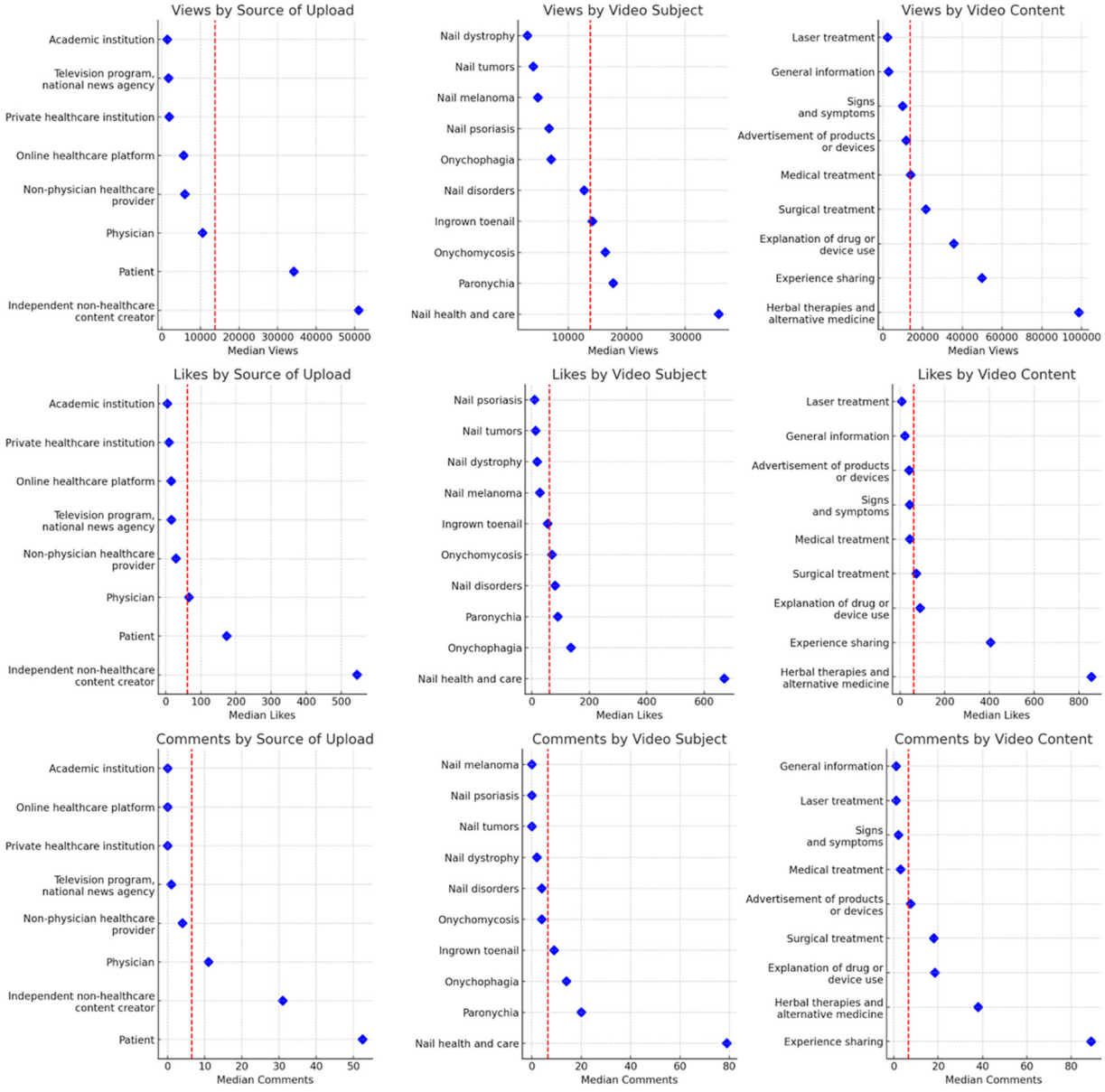

Figure 5. Median views, likes, and comments on YouTube videos are based on the source of upload, subject, and content. The red dotted line in each chart represents the overall median value across all categories for each metric, providing a benchmark to compare different groups

The quality of videos also varied depending on the individual providing the information in the video. Content creators who are not healthcare providers had the lowest GQS scores, with a median of 1.0, reflecting the poorest quality among the groups (Table 3). Non-physician healthcare providers had a median GQS of 3.0, while physicians (excluding dermatologists) also had a median score of 3.0. However, dermatologists had the highest overall quality, with a median GQS of 4.0. The differences in GQS scores across the groups were statistically significant (p<0.001), highlighting the superior quality of videos created by dermatologists. Post-hoc analysis confirmed that no statistical difference was found between non-physician healthcare providers and physicians from other specialties than dermatologists (p>0.05).

Finally, videos discussing or explaining surgical treatments were analyzed according to whether the information provider in the video was a dermatologist or non-dermatologist physician. Only 11 videos (2.2% of all videos) in which dermatologists gave opinions about surgical treatments were identified, whereas other physicians were featured in 58 (11.6%) surgical videos. Only one video described the nail biopsy procedure, which is produced and uploaded by a dermatologist. In parallel with other findings, the number of views, likes, and comments was higher in non-dermatologists, while GQS scores were higher in dermatologists (Figure 7, see plots for p-values).

Table 2. Comparison of median views, likes, and comments between individuals providing information in the YouTube videos

Informant in the video	Number of views		Number of likes		Number of comments	
	Median	p-value	Median	p-value	Median	p-value
Between health professionals and others						
Healthcare providers	5,529	**<0.001**	28	**<0.001**	2	**<0.001**
Informants other than healthcare providers	46,436		399		42	
Among health professionals						
Physicians	6,714	**0.015**	36	**0.017**	2	**0.369**
Non-physician healthcare providers	1,931		15.5		2	
Among physicians						
Dermatologists	1,678.5	**<0.001**	9	**<0.001**	0	**<0.001**
Physicians of other specialties	13,102		75		11	

Table 3. Distribution and comparison of the GQS scores across different categories of individuals providing information in YouTube videos

Informant in the video	Total n	Number of videos for each GQS					Overall GQS		
		1	2	3	4	5	Mean	Median	p-value
Content creators other than healthcare providers	165	86	49	21	8	1	1.7	1.0	**<0.001**
Non-physician healthcare providers	104	20	22	24	29	9	2.9	3.0*	
Physicians other than dermatologists	153	10	40	36	48	19	3.2	3.0*	
Dermatologists	78	2	9	17	24	26	3.8	4.0	

*Post-hoc analysis showed no statistical difference between subgroups (p>0.05).
GQS: Global Quality Scale

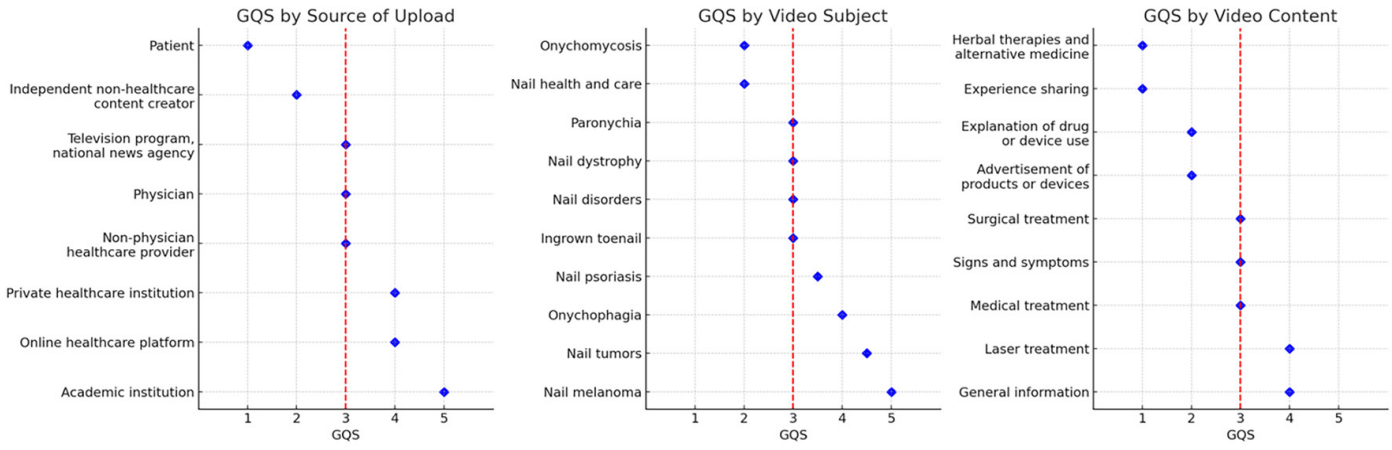

Figure 6. Comparison of the GQS of YouTube videos by source of upload, subject, and content. The red dotted line in each chart represents the overall median value

GQS: Global Quality Scale

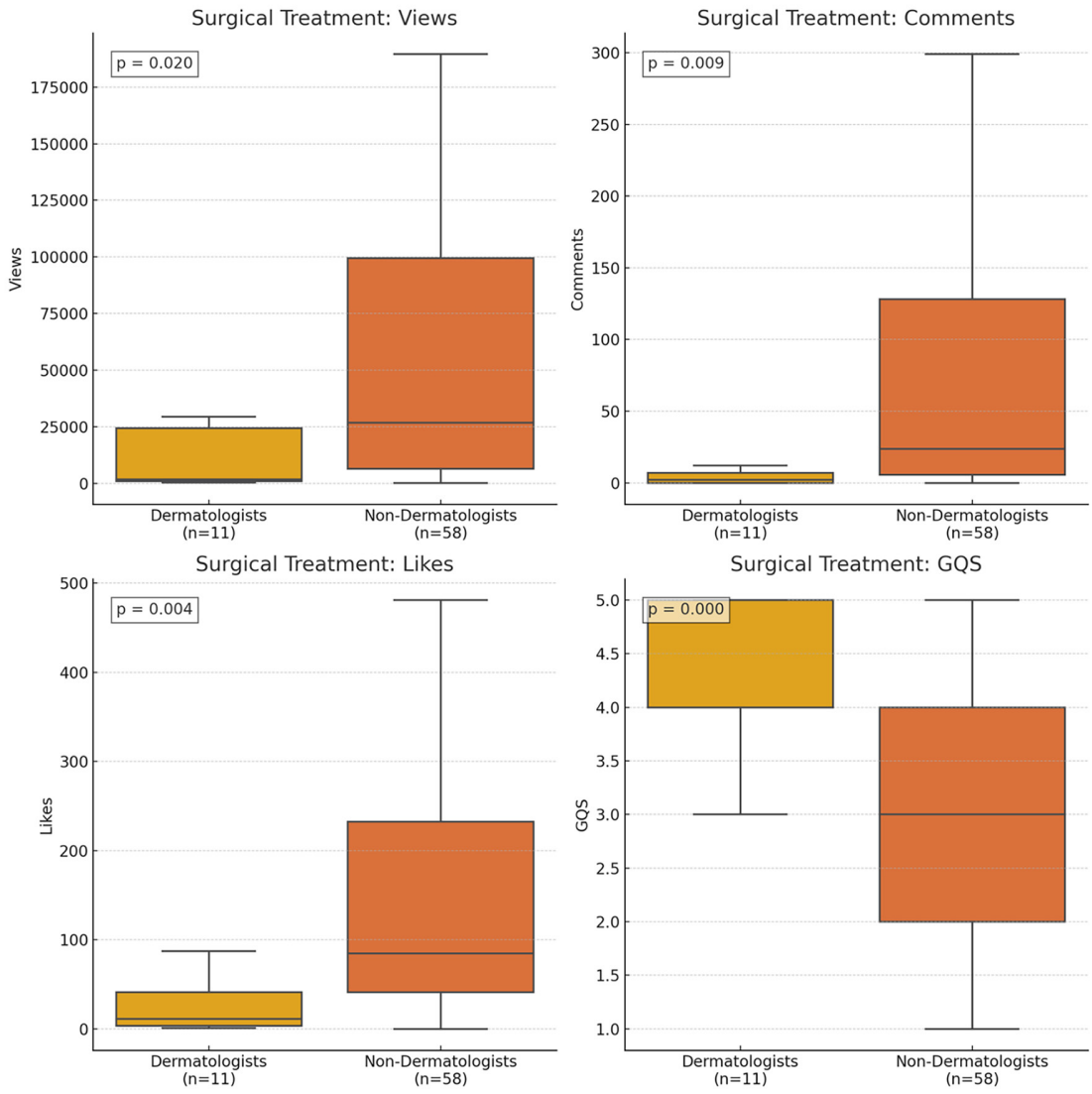

Figure 7. Comparison of views, likes, comments, and GQS between dermatologists and non-dermatologist physicians for videos containing surgical treatments

GQS: Global Quality Scale

Discussion

This study comprehensively evaluates Turkish YouTube content on nail health and disorders, revealing key trends in video topics, interactions, and quality. We found that ingrown toenails and onychomycosis had the most content produced, while videos about nail health and care received the highest levels of interaction. Although most videos provided general information on various conditions or covered surgical treatments, herbal and alternative therapies attracted the most attention from viewers. The highest GQS scores were given to videos on serious topics like nail melanoma and tumors, but these videos had the fewest views, likes, and comments. Non-healthcare content creators and patients dominated engagement, even though their videos were rated lower in quality. In contrast, healthcare professionals, particularly dermatologists, produced higher-quality content but received less interaction. Similarly, dermatologists had higher GQS scores when discussing surgical treatments than other specialists but still received less engagement.

The discrepancy between the video quality and audience engagement in health-related YouTube videos has been documented in numerous studies. For example, a study by Kaya and Sarıkaya Solak[12] from Türkiye, evaluating YouTube content on acne treatment, found that videos created by non-physicians garnered significantly more views, likes, and comments than physicians. However, the DISCERN score, which assesses the quality and reliability of health information, and the GQS score were notably higher for videos produced by physicians, particularly dermatologists. Despite the superior quality of these physician-created videos, they consistently attracted lower engagement, reflecting a pattern also observed in our study. Dermatologists were found to provide more comprehensive information, prioritizing evidence-

based medical treatments, whereas non-physicians often highlighted alternative therapies such as platelet-rich plasma, chemical peelings, and laser treatments.

Similarly, a study conducted by Güder and Güder[13] from Türkiye analyzed YouTube content related to psoriasis treatment and found that a significant portion of the videos was produced by non-physician content creators, including pharmacists, herbalists, and patients. These creators often promoted non-evidence-based treatment options, with herbal remedies featured in 65.7% of the videos. Despite the lower quality of these videos, they achieved high levels of engagement, paralleling the trend observed in our study. Güder and Güder[13] research also highlighted that videos created by healthcare professionals, particularly dermatologists, offered more comprehensive and evidence-based information. In line with Güder and Güder[13] findings, a study from the U.S. by Pithadia et al.[8] also reported that over half of the YouTube videos (55.3%) on topical psoriasis treatments, out of the 199 videos they analyzed, promoted natural remedies and alternative medicine rather than evidence-based therapies.

In the existing literature, only two studies have specifically analyzed YouTube content related to nail disorders: one by Nickles et al.[14], which focused on onychomycosis treatment, and the other by Ishack and Lipner[15], which examined nail biopsy procedures. Consistent with our findings, Nickles et al.[14] reported that general informative content was more common in onychomycosis treatment videos, but videos featuring patient experiences were more prevalent among viewers. These patient-driven videos frequently recommended natural remedies such as tea tree oil, apple cider vinegar, and bleach. In the study by Ishack and Lipner[15], the overall quality of videos on nail biopsy procedures was found to be low, with a mean DISCERN score of 1.60 out of 5, reflecting poor information. They also identified significant gaps in essential details, such as anesthetic techniques and repair methods, underscoring the need for higher-quality educational content for patients and physicians. In our study, there was also a noticeable absence of Turkish YouTube content addressing biopsy techniques, highlighting an area needing improvement.

The popularity of non-scientific content, mainly herbal and alternative treatments, presents risks for patients looking for accurate health information. Misinformation can lead to delayed or inappropriate treatments, which is especially dangerous for conditions like melanoma or nail tumors, where early and accurate diagnosis is vital. To address the gap between the quality of information and audience engagement, healthcare professionals need to adapt their communication strategies. This could include simplifying medical jargon and creating more visually appealing, easy-to-understand videos that connect with a broader audience. Academic institutions, professional organizations, and dermatologists themselves should also take a more active role in promoting evidence-based content by endorsing reliable videos on platforms like YouTube. These steps help ensure that accurate and trustworthy health information reaches more people.

Study Limitations

A key limitation of this study is its focus on Turkish-language YouTube content, which may limit the generalizability of the findings to other languages or regions. Another limitation is using a single scale (GQS) to assess video quality, which may not capture all aspects of content quality. However, the study's strength lies in its comprehensive evaluation of a large dataset over a broad time range, providing valuable insights into Turkish content's quality and engagement trends on nail health and disorders.

Conclusion

This study reveals that Turkish YouTube content on nail health and disorders is heavily dominated by non-healthcare content creators, who, despite providing lower-quality information, garnered the highest levels of engagement. Conversely, healthcare professionals, primarily dermatologists, produced higher-quality videos but reached fewer viewers. These results highlight the challenge of closing the gap between engagement and accurate, evidence-based information, as many popular videos could mislead patients. Efforts should be made to enhance the online presence of healthcare professionals and improve public access to accurate, high-quality health content to prevent the spread of misinformation.

Ethics

Informed Consent: Ethics committee approval was not required, as the study did not involve human or animal participants, and the videos were publicly accessible.

Footnotes

Authorship Contributions

Surgical and Medical Practices: O.E., V.A.E., Concept: O.E., V.A.E., A.S.Ş., M.S.G., Design: O.E., V.A.E., M.S.G., Data Collection or Processing: O.E., V.A.E., E.E.A., S.T., A.S.Ş., P.G.D., Analysis or Interpretation: O.E., V.A.E., M.S.G., Literature Search: O.E., V.A.E., Writing: O.E., V.A.E., M.S.G.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Centers for Disease C, Prevention: Health information technology use among adults: United States, 2022. CDC National Center for Health Statistics. 2023:1-5.
2. Madathil KC, Rivera-Rodriguez AJ, Greenstein JS, Gramopadhye AK: Healthcare information on YouTube: A systematic review. Health Informatics J. 2014;21:173-94.
3. Osman W, Mohamed F, Elhassan M, Shoufan A: Is YouTube a reliable source of health-related information? A systematic review. BMC Med Educ. 2022;22:382.
4. Nickles MA, Rustad AM, Ogbuefi N, McKenney JE, Stout M: What's being recommended to patients on social media? A cross-sectional analysis of acne treatments on YouTube. J Am Acad Dermatol. 2022;86:920-3.
5. Thang CJ, Garate D, Thang J, Lipoff JB, Barbieri JS: Short-form medical media: A multi-platform analysis of acne treatment information in TikTok videos, Instagram reels, and YouTube shorts. JMIR Dermatol. 2023;6:e48140.
6. Mueller SM, Hongler VNS, Jungo P, et al: Fiction, falsehoods, and few facts: Cross-sectional study on the content-related quality of atopic eczema-related videos on YouTube. J Med Internet Res. 2020;22:e15599.
7. Freemyer B, Drozd B, Suarez A: A cross-sectional study of YouTube videos about atopic dermatitis. J Am Acad Dermatol. 2018;78:612-3.
8. Pithadia DJ, Reynolds KA, Lee EB, Wu JJ: A cross-sectional study of YouTube videos as a source of patient information about topical psoriasis therapies. J Dermatolog Treat. 2020; 31:366-9.

9. Pithadia DJ, Reynolds KA, Lee EB, et al: A cross-sectional study of YouTube videos as a source of patient information about phototherapy and excimer laser for psoriasis. J Dermatolog Treat. 2020;31:707-10.
10. Raime DP, Ezaldein N, Quiñones M, Chow K, Lukas K, Ezaldein HH: YouTube as a source of Hidradenitis suppurativa patient education: A social media content analysis. Cureus. 2023;15:e38047.
11. Killion L, Therianou A, Watchorn RE: What's being recommended to patients on social media? A cross-sectional analysis of alopecia treatments on YouTube(®). Clin Exp Dermatol. 2022;47:759-63.
12. Kaya Ö, Sarıkaya Solak S: Evaluation of the quality, reliability, and popularity of Turkish YouTube videos on acne treatment. Turkderm-Turk Arch Dermatol Venereol. 2022;56:166-71.
13. Güder H, Güder S: Psoriasis treatment on YouTube: a cross-sectional study. Turkiye Klinikleri Journal of Health. 2023;13:101230.
14. Nickles MA, Pavelka M, Mervak JE: Onychomycosis on YouTube: A cross-sectional analysis. Skin Appendage Disord. 2022;8:307-11.
15. Ishack S, Lipner SR: Evaluating the impact and educational value of YouTube videos on nail biopsy procedures. Cutis. 2020;105:148-9;e1.
16. Bernard A, Langille M, Hughes S, et al: A systematic review of patient inflammatory bowel disease information resources on the World Wide Web. Am J Gastroenterol. 2007;102:2070-7.

Original Investigation
Orijinal Araştırma

DOI: 10.4274/turkderm.galenos.2025.44969
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):54-9

Retrospective evaluation of 28 cases of inflammatory linear verrucous epidermal nevus

Enflamatuvar lineer verrüköz epidermal nevüs: 28 olgunun retrospektif değerlendirmesi

Buğra Burç Dağtaş*, Vildan Manav*, Cem Leblebici, Ayşe Esra Koku Aksu***

*University of Health Sciences Türkiye, İstanbul Training and Research Hospital, Department of Dermatology, İstanbul, Türkiye
**University of Health Sciences Türkiye, İstanbul Training and Research Hospital, Department of Pathology, İstanbul, Türkiye

Abstract

Background and Design: Inflammatory linear verrucous epidermal nevus (ILVEN) is a rare, chronic dermatosis characterized by linear erythematous scaly plaques following Blaschko's lines. This study aims to comprehensively evaluate ILVEN cases, focusing on demographic, clinical, and histopathological features to improve understanding of its presentation and management.
Materials and Methods: This retrospective cross-sectional study included 28 patients diagnosed at our center with clinically and histopathologically confirmed ILVEN between 2011 and 2022. Data on patient demographics, age at lesion onset, duration before diagnosis, lesion location, histopathological findings, and treatment approaches were collected from hospital records and supplemented by patient interviews.
Results: Our study had a male predominance (63.4%), with a median age of lesion onset at 7.5 years and a median age of 14 years, indicating a prolonged delay in diagnosis. The mean duration from lesion onset to diagnosis was 9.86 years. Lesions were most frequently located on the lower extremities, following a Blaschkoid distribution, with 73.3% of patients reporting pruritus. Key histopathological findings included orthokeratosis with hypergranulosis (60.7%), parakeratosis overlying hypogranulosis (46.4%), and psoriasiform epidermal hyperplasia (46.4%). Topical corticosteroids were the primary treatment modality, while spontaneous regression was observed in 14.3% of untreated cases.
Conclusion: This study highlights the clinical and histopathological features of ILVEN, emphasizing the importance of early recognition in reducing diagnostic delays. Further research is warranted to enhance understanding of ILVEN's clinical course and to optimize diagnostic and therapeutic strategies.
Keywords: ILVEN, Epidermal nevus, Linear dermatosis

Öz

Amaç: Enflamatuvar lineer verrüköz epidermal nevüs (ILVEN), Blaschko çizgilerini izleyen lineer eritematöz, skuamlı plaklarla karakterize nadir ve kronik bir dermatozdur. Bu çalışma, ILVEN'in sunumu ve yönetimi hakkında daha fazla bilgi sağlamak amacıyla olguları demografik, klinik ve histopatolojik özellikler açısından kapsamlı bir şekilde değerlendirmeyi amaçlamaktadır.
Gereç ve Yöntem: Bu retrospektif kesitsel çalışmaya, 2011-2022 yılları arasında merkezimizde klinik ve histopatolojik olarak ILVEN tanısı doğrulanmış 28 hasta dahil edilmiştir. Hastaların demografik verileri, lezyon başlangıç yaşı, tanıya kadar geçen süre, lezyon yerleşimi, histopatolojik bulgular ve tedavi yaklaşımlarına ilişkin veriler hastane kayıtlarından toplanmış ve hasta görüşmeleri ile desteklenmiştir.
Bulgular: Çalışmamızda erkek hastalar çoğunluktaydı (%63,4) ve lezyon başlangıç yaşı ortancası 7,5 yıl, tanı yaşı ortancası 14 yıl olarak bulunmuş olup, tanıda belirgin bir gecikme olduğunu göstermektedir. Lezyon başlangıcından tanıya kadar geçen ortalama süre 9,86 yıl olarak saptanmıştır. Lezyonlar en sık alt ekstremitelerde, Blaschkoid dağılımda bulunmuş ve hastaların %73,3'ü kaşıntı bildirmiştir. Ana histopatolojik

Address for Correspondence/Yazışma Adresi: Buğra Burç Dağtaş MD, University of Health Sciences Türkiye, İstanbul Training and Research Hospital, Department of Dermatology, İstanbul, Türkiye
E-mail: bugradagtas@gmail.com **ORCID:** orcid.org/0000-0002-9592-9968
Received/Geliş Tarihi: 03.11.2024 **Accepted/Kabul Tarihi:** 23.03.2025 **Publication Date/Yayınlanma Tarihi:** 01.07.2025

Cite this article as/Atıf: Dağtaş BB, Manav V, Leblebici C, Koku Aksu AE. Retrospective evaluation of 28 cases of inflammatory linear verrucous epidermal nevus. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):54-9

bulgular hipergranülozis ile ortokeratoz (%60,7), hipogranülozis üzerinde parakeratoz (%46,4) ve psoriasiform epidermal hiperplazi (%46,4) şeklindedir. Ana tedavi yöntemi topikal kortikosteroidler olup, tedavi almayan hastaların %14,3'ünde spontan gerileme gözlenmiştir.
Sonuç: Bu çalışma, ILVEN'in klinik ve histopatolojik özelliklerini vurgulayarak erken tanının tanı gecikmelerini azaltmadaki önemine dikkat çekmektedir. ILVEN'in klinik seyrini daha iyi anlamak ve tanı ile tedavi stratejilerini optimize etmek için daha fazla araştırmaya ihtiyaç vardır.
Anahtar Kelimeler: İLVEN, Epidermal nevüs, Lineer dermatozlar

Introduction

Inflammatory linear verrucous epidermal nevus (ILVEN), first described by Altman and Mehregan[1], is a rare skin disorder characterized by a linear pattern of erythematous scaly papules and plaques of various sizes following Blaschko's lines. Although it is usually congenital or seen in early childhood, it can rarely be seen in adulthood[1-3]. It is thought to be more common in women than men, and most commonly involves the lower extremities[1,2]. Central nervous system and skeletal anomalies may also be observed. Rarely, oral and mucosal lesions have been reported[4]. The classic histology of ILVEN is psoriasiform acanthosis with overlying alternating parakeratosis and orthokeratosis and corresponding hypo- and hypergranulosis underneath[5]. The differential diagnosis should consider disorders with similar distribution patterns, such as non-inflammatory epidermal nevus, linear psoriasis, linear lichen planus, and lichen striatus[6]. Although multiple case reports on ILVEN are available, there remains a notable lack of larger descriptive studies that can provide a broader understanding of its demographic, clinical, and histopathologic characteristics of ILVEN.

In this study, we aimed to comprehensively assess patients' demographic, clinical, and histopathologic characteristics with a confirmed clinical and histopathologic diagnosis of ILVEN. Additionally, we sought to explore whether specific demographic or clinical features in our cohort align with or differ from those reported in existing literature, with particular attention to possible gender-related differences.

Material and Methods

Study Design

This study was a retrospective, cross-sectional analysis conducted at our dermatology department. It included patients diagnosed with ILVEN between 2011 and 2022.

Setting and Participants

Eligible participants were identified through a rigorous review of medical records within the hospital's information system. Inclusion criteria required both clinical and histopathological confirmation of ILVEN. Out of 66 patients who had undergone biopsy with a preliminary diagnosis of ILVEN, 28 met the inclusion criteria based on verified clinical and histopathological findings. Patients without conclusive diagnostic confirmation were excluded to ensure diagnostic precision. Missing or incomplete data were meticulously supplemented through follow-up telephone interviews with patients or, where applicable, their legal guardians to complete the dataset. Informed consent for photography was obtained from parents or legal guardians for minor patients, following ethical guidelines. Ethics committee approval was obtained for the study (approval number: 406, date: 23.12.2022).

Variables and Data Sources

For each patient, demographic data (gender, age at presentation, and age of lesion onset) and clinical characteristics (lesion duration, location, distribution pattern, and presence or absence of pruritus) were documented based on hospital records. Histopathological findings were recorded from biopsy reports, with all variables carefully corroborated by available clinical records and, where necessary, follow-up interview responses to ensure data completeness. Treatment modalities were classified by the primary intervention type, with spontaneous regression, if observed, also noted. The selected variables were based on established ILVEN characteristics in the literature to support a comprehensive analysis.

Bias

Potential sources of bias were meticulously addressed by rigorously validating hospital record data with interview-obtained information, particularly for variables where data completeness was crucial. Given the retrospective nature of data collection, there was a potential for recall bias, especially concerning lesion duration and age of onset; however, structured interview protocols and verification against hospital records were used to enhance reliability and accuracy in reported information, ensuring the highest level of objectivity in our study.

Statistical Analysis

All statistical analyses were performed using IBM SPSS Statistics version 24.0 (SPSS Inc., Chicago, IL, USA). Given the data's non-parametric distribution, continuous variables were summarized using descriptive statistics and presented as median values with minimum and maximum ranges. The normality of continuous variables was assessed via histograms and Q-Q plots. Continuous variables were compared between subgroups using the Mann-Whitney U test, and categorical variables were analyzed using Pearson's chi-square test. A significance threshold of $p<0.05$ was applied to all statistical tests.

Results

Participant Flow

The study initially identified 66 patients with a preliminary diagnosis of ILVEN; however, only 28 patients met the inclusion criteria after clinical and histopathological confirmation. All patients were included due to missing data, as any incomplete information was supplemented through follow-up interviews with the patients or their legal guardians.

Descriptive Data

The final cohort included 18 males (63.4%) and 10 females (36.6%), with a median age of 10 years at the time of presentation (Table 1). The mean age at lesion onset was 10.07±15.22 years, with a median of 7.5 years and a range from 0 to 77 years. No statistically significant difference in age of onset was observed between genders ($p=0.470$). The median duration of lesions before clinical presentation was 4.0 years (mean =9.86±12.32 years, range =0.2 to 48.0 years), with no significant gender differences ($p=0.104$). The mean age at diagnosis was 19.93±16.93 years, with a median of 14 years and a range of 2 to 78 years, also showing no significant gender-based differences ($p=0.259$).

Turkderm - Turk Arch Dermatol Venereol
2025;59(2):54-9

Clinical Characteristics and Outcome Data

Lesion location varied, with the most common sites being the lower extremities (12 patients, 42.9%), followed by the upper extremities (9 patients, 32.1%), head and neck (4 patients, 14.3%), and trunk (3 patients, 10.7%). Anogenital involvement was identified in one patient (3.6%) (Figure 1). Pruritus was reported by 73.3% of the patients, while 26.7% were asymptomatic. No associated congenital anomalies were detected in any of the patients. Most lesions (89%) followed a Blaschkoid distribution, 82% exhibited a verrucous texture, and 75% displayed scaling. Regarding lesion morphology, plaques were the predominant presentation (93%), while papules were observed in 7%. Representative clinical photographs are presented in Figures 2 and 3.

Differential diagnoses considered included non-inflammatory epidermal nevus (20 patients, 71.4%), linear psoriasis (15 patients, 53.6%), linear lichen planus (11 patients, 39.3%), lichen striatus (9 patients, 32.1%), and verruca plana (7 patients, 25%).

Histopathological examination revealed orthokeratosis with hypergranulosis in 60.7% of patients (17 patients), parakeratosis overlying hypogranulosis in 46.4% (13 patients), psoriasiform epidermal hyperplasia in 46.4% (13 patients), papillomatosis in 39.3% (11 patients), and acanthosis in 32.1% (9 patients) (Table 1, Figure 4).

Topical corticosteroids were the primary treatment modality for 39.3% of patients, followed by a combination of topical corticosteroids and emollients (17.9%). Cryotherapy and laser therapy were each applied in 3.6% of cases. Spontaneous regression occurred in 14.3% of untreated patients, observed during follow-up (Table 1).

Discussion

Key Results

This study provides a detailed evaluation of the demographic, clinical, and histopathological characteristics of patients with confirmed ILVEN. Our study indicates a male predominance, with 63.4% of patients being male. This finding contrasts with earlier studies that reported a higher prevalence of ILVEN in females. For example, Altman and Mehregan[1], reported a 4:1 female-to-male ratio in their cohort of 25 patients, suggesting that females were more commonly affected. However, our findings are consistent with those of Lee and Rogers[2] who found that 70% of their 23 patients were male. The variability in gender distribution across studies may be attributed to differences in sample size, population demographics, or even regional variations in the disease's presentation. It is important to note that the reasons for this observed gender difference remain unclear. While earlier studies suggested hormonal or genetic factors might play a role, no definitive explanations have been established. Further large-scale studies are

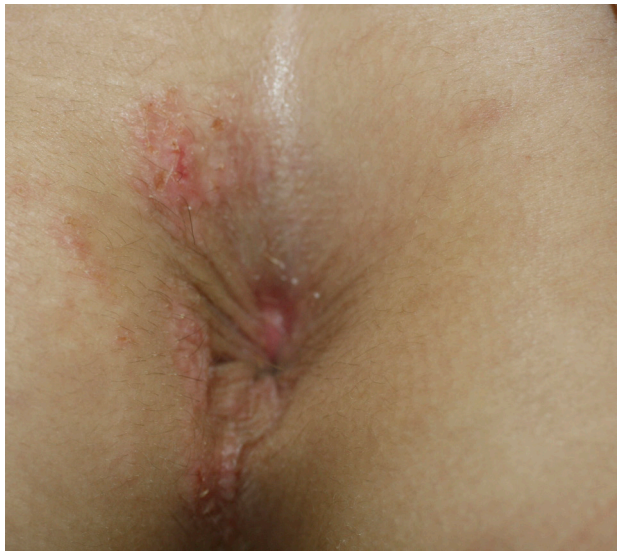

Figure 1. Scattered plaque in the anogenital area is verrucous in places and compatible with ILVEN, in which scale is seen

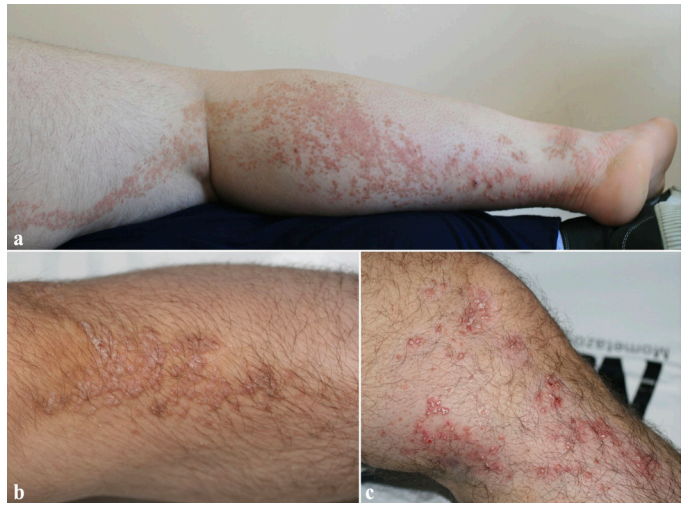

Figure 2. (a) Plaque with blaschkoid distribution, erythematous, verrucous appearance extending from the posterior right thigh to the ankle. **(b)** Plaque in the right peripatellar area, consisting of many papules with a linear distribution, verrucous appearance, and pinkish color, tends to merge. **(c)** Erythematous scaly plaque with blaschkoid distribution and verrucous appearance extending from the left peripatellar region to the center of the cruris

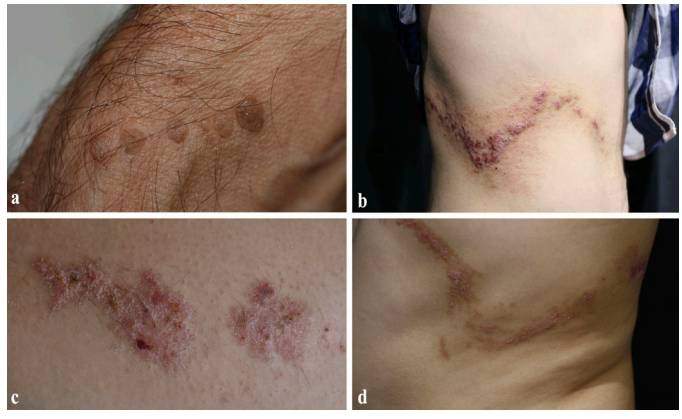

Figure 3. (a) Plaque consisting of linearly distributed, verrucous, brownish papules in the flexor region of the right forearm. **(b)** Erythematous scaly plaque with blaschkoid distribution, verrucous appearance on the left side of the trunk. **(c)** Linearly distributed, verrucous, erythematous plaque with a mild scaly appearance on the posterior thigh. **(d)** A mildly erythematous brownish plaque with blaschkoid distribution and a verrucous appearance is located on the left side of the trunk

needed to explore whether these gender differences are consistent across diverse populations and what factors may contribute to ILVEN's variable presentation between males and females.

The broad age range at diagnosis and prolonged delay in clinical presentation observed in our study align with the indolent progression of ILVEN reported in prior studies. Altman and Mehregan[1], found that while ILVEN often presents before age five, the mean duration of lesions before diagnosis was approximately six years, highlighting this condition's persistence and often overlooked nature.

Our study observed a slightly higher mean age of onset and an extended diagnostic timeline, including one unique case presenting at 77. Although ILVEN generally manifests in childhood, cases with late-onset presentation have also been reported in the literature, suggesting that ILVEN may occasionally emerge in later decades of life[3,7,8].

Table 1. Characteristics of ILVEN Patients (n=28)

Characteristic	Frequency (n, %)		Mean ± SD	Median (Range)
Demographic Information				
Gender	Male	63.4% (18)		
	Female	36.6% (10)		
Clinical Information				
Age at lesion onset (years)			10.07±15.22	7.5 (0-77)
Duration of lesions (years)			9.86±12.32	4.0 (0.2-48.0)
Age at diagnosis (years)			19.93±16.93	14 (2.0-78.0)
Location of Lesions	Lower extremity	42.9% (12)		
	Upper extremity	31.1% (9)		
	Head and neck	14.3% (4)		
	Trunk	10.7% (3)		
	Anogenital region	3.6% (1)		
Symptoms	Itching	73.3% (20)		
	Asymptomatic	26.7% (8)		
Distribution of Lesions	Blaschkoid	89% (25)		
	Verrucous appearance	82% (23)		
	Scaling	75% (21)		
Appearance of Lesions	Plaque	90% (26)		
	Papule	10% (2)		
Histopathological Findings	Orthokeratosis with hypergranulosis	60.7% (17)		
	Parakeratosis over hypogranulosis	46.4% (13)		
	Psoriasiform epidermal hyperplasia	46.4% (13)		
	Papillomatosis	39.3% (11)		
	Acanthosis	32.1% (9)		
Differential Diagnosis	Non-inflammatory epidermal nevus	71.4% (20)		
	Linear psoriasis	53.6% (15)		
	Linear lichen planus	39.3% (11)		
	Lichen striatus	32.1% (9)		
	Verruca plana	25% (7)		
Treatment Methods	Topical corticosteroids	39.3% (11)		
	Topical corticosteroids and emollient	17.9% (5)		
	Cryotherapy	3.6% (1)		
	Laser Therapy	3.6% (1)		
	Spontaneous regression	14.3% (4)		
Other Findings	Associated anomalies	None		
	Family history	None		

ILVEN: Inflammatory linear verrucous epidermal nevus, SD: Standard deviation.

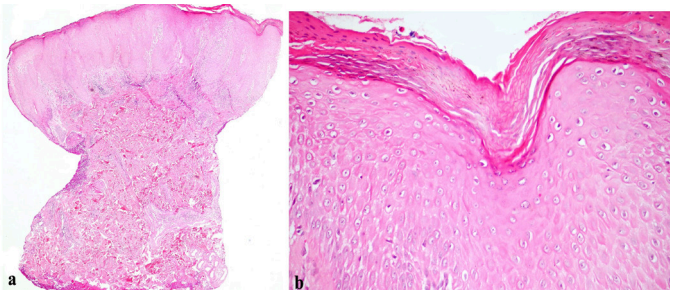

Figure 4. (a) Psoriasiform epidermal hyperplasia (HE x 100) **(b)** Alternating orthohyperkeratosis and parakeratosis areas are observed. The granular layer is reduced/absent in the parakeratosis area. The granular layer can be seen in ortho-hyperkeratosis areas (HE x 400)
HE: Hematoxylin and Eosin staining

These findings underscore the need for prospective studies to elucidate ILVEN's natural history across populations and emphasize the importance of increased clinical awareness to promote earlier diagnosis.

Limitations

This study has several limitations that should be considered when interpreting the results. The study's retrospective nature may introduce recall and selection biases, particularly concerning the age of onset and lesion duration, as some data were supplemented through telephone interviews with patients or their guardians. The relatively small sample size may limit the generalizability of the findings and reduce the statistical power to detect potential differences in demographics or clinical features. As this is a single-center study, findings may reflect local or regional patterns that need to be more generalizable to broader populations. Future studies with more significant, multicenter cohorts must confirm our observations and establish more generalizable findings about ILVEN's demographic and clinical characteristics.

Interpretation

The clinical presentation of ILVEN in our cohort was consistent with previously documented findings. Most lesions followed a Blaschkoid distribution and exhibited verrucous texture and scaling. Plaque morphology was the predominant form, with papules observed less frequently. This aligns with prior studies identifying plaques as characteristic of ILVEN and often forming linear patterns along Blaschko's lines[9,10].

Additionally, 73.3% of our patients reported itching associated with their ILVEN lesions, consistent with the literature, where pruritus is frequently noted as a prominent symptom. Altman and Mehregan[1], initially described ILVEN as often accompanied by pruritus, which can be persistent and resistant to treatment, though some patients may remain asymptomatic[1].

These findings emphasize the typical clinical presentation of ILVEN and the variability in symptom severity, underscoring the importance of individualized management strategies, especially for pruritic cases where standard treatments may offer only temporary relief.

Differentiating ILVEN from other linear dermatoses is essential due to overlapping clinical presentations but distinct histopathological and etiological features.

The histopathological findings in our cohort are consistent with the established patterns in ILVEN, as reported in the literature. Psoriasiform epidermal hyperplasia and alternating orthokeratosis with hypergranulosis and parakeratosis overlying hypogranulosis were among the most common features observed, aligning with descriptions by Altman and Mehregan[1], in their foundational study on ILVEN of orthokeratosis with a thickened granular layer and parakeratosis with a diminished granular layer is characteristic of ILVEN. It is often essential for distinguishing it from other linear dermatoses[1]. Recent studies have further refined the diagnostic approach with immunohistochemical markers, showing lower basal layer Ki-67 positivity and increased keratin-10 expression in ILVEN compared to psoriasis, along with decreased and localized involucrin staining in parakeratotic regions[3]. Additionally, recent studies suggest a genetic basis involving CARD14 mutations in some cases, highlighting the potential role of mosaicism in ILVEN's variable presentation[5]. Given these findings, further genetic studies are warranted to understand ILVEN's pathogenesis better and refine diagnostic and therapeutic approaches.

Our study's most common differential diagnosis was non-inflammatory epidermal nevus, which, although similar in its verrucous appearance, is typically associated with congenital anomalies and lacks the lower extremity predominance seen in ILVEN[11]. The absence of accompanying anomalies in our cases further corroborated the diagnosis of ILVEN, distinguishing it from non-inflammatory epidermal nevus and similar entities.

Linear psoriasis, another frequently considered differential diagnosis, is often adult-onset and shares morphological similarities with ILVEN. However, it exhibits distinct immunohistochemical features, with studies like those by Vissers et al.[12] showing lower Ki-67 positivity, higher keratin-10 positivity, and Human Leukocyte Antigen-DR isotype expression in ILVEN.. In our study, many patients required differentiation from linear psoriasis, and the characteristic alternating ortho- and parakeratosis observed in ILVEN were vital in confirming the diagnosis.

Although linear lichen planus may present in a linear pattern similar to ILVEN, it is characterized by violaceous papules and plaques, later onset, and more intense pruritus. Histopathologically, it also exhibits a band-like lymphocytic infiltrate, differentiating it from ILVEN[13]. In our cohort, ILVEN's earlier onset and Blaschkoid distribution helped to distinguish it from linear lichen planus.

Lichen striatus, which often affects children, is characterized by its spontaneous regression and unique histopathological features, such as lymphocytic infiltrates around sweat ducts[14]. Although a minority of ILVEN cases in our cohort demonstrated spontaneous regression, the persistent Blaschkoid distribution and absence of lichen striatus-specific histopathology were decisive factors favoring ILVEN.

Verruca plana, associated with human papillomavirus (HPV) infection, presents as flat, verrucous, hyperkeratotic lesions and can be differentiated from ILVEN by the histological presence of koilocytes, detection of HPV DNA via PCR, and dermoscopic differences[15].

A range of treatments has been employed in ILVEN cases with varying degrees of success, including topical and intralesional corticosteroids, retinoids, dithranol, 5-fluorouracil, vitamin D analogs, calcineurin inhibitors, surgical excision, cryotherapy, and laser therapies[16,17]. Recently, biological agents have also shown promising results[18].

In our study, topical corticosteroids were the primary treatment modality (39.3%), followed by combinations of corticosteroids and emollients (17.9%), with cryotherapy and laser therapy used less frequently. These interventions align with standard management strategies but typically provide only temporary relief, as ILVEN generally exhibits limited responsiveness to treatment. Notably, spontaneous resolution occurred in 14.3% of untreated patients-a phenomenon occasionally reported in the literature that warrants further investigation to identify potential predictors. Given these findings, further genetic and immunohistochemical research is recommended to optimize treatment protocols and elucidate the factors driving ILVEN's variable clinical course.

Generalisability

Our findings offer valuable insights into ILVEN's clinical and histopathological characteristics; however, the single-center design and the regional focus of the sample may limit their applicability to other populations. Future studies involving broader, more diverse populations across various geographic areas are necessary to enhance the external validity of these results and provide a more comprehensive understanding of ILVEN's demographic variability.

Conclusion

In this study, we evaluated the demographic, clinical, and histopathological characteristics of patients diagnosed with ILVEN, a rare variant of epidermal nevus. Recognizing the clinical and histopathological features of ILVEN is crucial for differentiating it from other linearly distributed dermatological conditions. Although ILVEN typically appears early, patients often delay seeking dermatological care, underscoring the importance of early detection and intervention. Further research is warranted to enhance understanding of ILVEN's clinical course and to optimize diagnostic and therapeutic strategies.

Ethics

Ethics Committee Approval: This study was approved by the ethics board of the University of Health Sciences Türkiye. İstanbul Training and Research Hospital (approval number: 406, date: 23.12.2022).

Informed Consent: Informed consent for photography was obtained from parents or legal guardians for minor patients, following ethical guidelines.

Footnotes

Authorship Contributions

Surgical and Medical Practices: B.B.D., V.M., C.L., A.E.K.A., Concept: B.B.D., V.M., Design: B.B.D., V.M., Data Collection or Processing: B.B.D., V.M., Analysis or Interpretation: B.B.D., A.E.K.A., Literature Search: B.B.D., Writing: B.B.D., A.E.K.A.

Conflict of Interest: The authors declared that they have no conflict of interest.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Altman J, Mehregan AH: Inflammatory linear verrucose epidermal nevus. Arch Dermatol. 1971;104:385-9.
2. Lee SH, Rogers M: Inflammatory linear verrucous epidermal naevi: a review of 23 cases. Australas J Dermatol. 2001;42:252-6.
3. Tseng HW, Liao JB, Wei YA: Adult-onset inflammatory linear verrucous epidermal nevus: Immunohistochemical studies and review of the literature. J Cutan Pathol. 2021;48:140-6.
4. Ozçelik D, Parlak AH, Oztürk A, Kavak A, Celikel N: Unilateral linear verrucous epidermal nevus of the face and the oral mucosa. Plast Reconstr Surg. 2005; 115:17e-9e.
5. Atzmony L, Ugwu N, Hamilton C, et al.: Inflammatory linear verrucous epidermal nevus (ILVEN) encompasses a spectrum of inflammatory mosaic disorders. Pediatr Dermatol. 2022;39:903-7.
6. Behera B, Devi B, Nayak BB, Sahu B, Singh B, Puhan MR: Giant inflammatory linear verrucous epidermal nevus: successfully treated with full thickness excision and skin grafting. Indian J Dermatol. 2013;58:461-3.
7. Kawaguchi H, Takeuchi M, Ono H, Nakajima H: Adult onset of inflammatory linear verrucous epidermal nevus. J Dermatol. 1999;26:599-602.
8. Kim R, Marmon S, Kaplan J, Kamino H, Pomeranz MK. Verrucous epidermal nevus. Dermatol Online J. 2013;19:20707.
9. Khachemoune A, Janjua SA, Guldbakke KK. Inflammatory linear verrucous epidermal nevus: a case report and short review of the literature. Cutis. 2006;78:261-7.
10. Rogers M: Epidermal nevi and the epidermal nevus syndromes: a review of 233 cases. Pediatr Dermatol. 1992; 9: 342-4.
11. Sugarman JL: Epidermal nevus syndromes. Semin Cutan Med Surg. 2004; 23:145-57.
12. Vissers WH, Muys L, Erp PE, de Jong EM, van de Kerkhof PC: Immunohistochemical differentiation between inflammatory linear verrucous epidermal nevus (ILVEN) and psoriasis. Eur J Dermatol. 2004;14: 216-20.
13. Wagner G, Rose C, Sachse MM: Clinical variants of lichen planus. J Dtsch Dermatol Ges. 2013;11:309-19.
14. Peramiquel L, Baselga E, Dalmau J, Roé E, del Mar Campos M, Alomar A: Lichen striatus: clinical and epidemiological review of 23 cases. Eur J Pediatr. 2006;165:267-9.
15. Rintala MA, Grénman SE, Järvenkylä ME, Syrjänen KJ, Syrjänen SM: High-risk types of human papillomavirus (HPV) DNA in oral and genital mucosa of infants during their first 3 years of life: experience from the Finnish HPV Family Study. Clin Infect Dis. 2005;41:1728-33.
16. Sood S, Bestavros S, Yilmaz O, et al.: Systemic Treatment Options for Inflammatory Linear Verrucous Epidermal Nevi: An Evidence-Based Review. J Cutan Med Surg. 2025;29:93-5.
17. Gokalp H, Armutlu A: Vulvar Inflammatory Linear Verrucous Epidermal Nevus: Remission with Nonsteroidal Antiinflammatory Cream. Turkiye Klinikleri Journal of Dermatology. 2017;27.
18. Grgurich E, Gupta N, Owen R, Purcell SM: Inflammatory linear verrucous epidermal nevus responsive to 308-nm excimer laser treatment. Cutis. 2018;102:111-4.

Case Report
Olgu Sunumu

DOI: 10.4274/turkderm.galenos.2025.42800
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):60-2

Atrichia congenita with papular lesions: A rare cause of pediatric alopecia

Papüler lezyonlu konjenital atrişi: Pediatrik alopesinin nadir bir nedeni

Bhumesh Kumar Katakam*, Arunima Dhabal, Narsimha Rao Netha Gurram*, Sudharani Chintagunta***

*Gandhi Medical College and Hospital, Department of Dermatology, Secunderabad, India
**Jagannath Gupta Institute of Medical Sciences and Hospital, Department of Dermatology, Kolkata,India

Abstract

Congenital atrichia with papules is a rare inherited disorder characterized by the replacement of hair follicles with keratinous cysts, resulting in hair loss shortly after birth and papular lesions. It should be differentiated from other causes of congenital atrichia, such as vitamin D-dependent rickets, alopecia universalis, and ectodermal dysplasia, to avoid unnecessary administration of medications and to counsel parents regarding the benign but irreversible nature of the condition.
Keywords: Congenital alopecia, atrichia, papular

Öz

Papüler lezyonlu konjenital atrişi, saç foliküllerinin yerini keratinöz kistlerin alması, doğumdan kısa süre sonra gelişen yaygın saç dökülmesi ve papüler deri lezyonları ile karakterize, nadir görülen kalıtsal bir hastalıktır. Gereksiz tedavilerden kaçınmak ve ebeveynleri hastalığın benign ancak irreversibl olduğu konusunda bilgilendirmek amacıyla; bu durumun D vitaminine bağımlı raşitizm, alopesi üniversalis ve ektodermal displazi gibi diğer konjenital atrişi nedenlerinden ayırt edilmesi gerekir.
Anahtar Kelimeler: Konjenital alopesi, atrişi, papüler

Introduction

Atrichia congenita with papular lesions (APL) is a rare inherited disorder caused by mutations in the human hairless gene (HR) on chromosome 8p21-22[1]. It is clinically characterized by the irreversible loss of hair shortly after birth, associated with a diffuse papular eruption. Histologically, these papules represent follicular keratin-filled cysts formed due to an abnormal hair cycle[2]. Herein, we report a case of APL in an Indian girl.

Case Report

A six-year-old girl presented with complete absence of scalp and body hair along with multiple raised lesions involving the scalp, face, body, and limbs. The child exhibited normal hair growth at birth. Spontaneous hair loss began at the age of 2 months on the scalp and gradually progressed to her eyebrows and eyelashes. Multiple treatment attempts failed to induce hair regrowth. At the age of 4 years, the parents noticed the appearance of asymptomatic raised lesions on her scalp and face, which eventually spread to the rest of her body. She

Address for Correspondence/Yazışma Adresi: Arunima Dhabal MD, Jagannath Gupta Institute of Medical Sciences and Hospital, Department of Dermatology, Kolkata, India
E-mail: ardhabal@gmail.com **ORCID:** orcid.org/0000-0003-2553-6453
Received/Geliş Tarihi: 11.05.2024 **Accepted/Kabul Tarihi:** 08.01.2025 **Publication Date/Yayınlanma Tarihi:** 01.07.2025

Cite this article as/Atıf: Katakam BK, Dhabal A, Gurram NRN, Chintagunta S. Atrichia congenita with papular lesions: A rare cause of pediatric alopecia. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):60-2

had no history of atopy, reduced sweating, hearing difficulty, blurred vision, seizures, or bone pain. She was the second child of a second-degree consanguineous marriage, born via Caesarean delivery after an uneventful pregnancy. She had attained developmental milestones at the usual age. Neither her elder sister nor her parents had any similar complaints.

Examination revealed a complete absence of hair throughout the scalp and body (Figure 1 and 2). There were multiple skin-colored to hyperpigmented discrete papules, ranging from 1 to 5 mm in size, distributed symmetrically over the scalp, face, trunk, and extremities, sparing the palms and soles (Figure 2). The nails, teeth, and mucosa did not show any abnormalities. Her physical development was normal for her age at presentation. There were no bony deformities or dysmorphic features, except for mild lateral protrusion of the ears. Differential diagnoses of APL, vitamin D-dependent rickets (VDDR) type IIA, alopecia universalis, and hidrotic ectodermal dysplasia were considered.

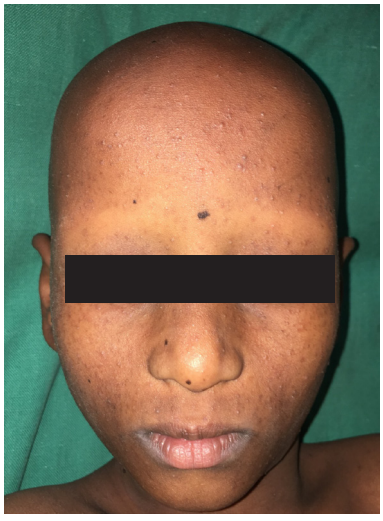

Figure 1. Complete loss of hair on scalp

Hematological, hepatic, and renal parameters, as well as calcium and phosphate levels, were within the normal range. The vitamin D3 level was 36.89 ng/mL (normal: 30-100 ng/mL). Radiographs of the bilateral wrist joints and lower limbs did not show any features suggestive of vitamin D deficiency. Thus, the clinical picture pointed towards a diagnosis of APL. However, histopathological evidence could not be obtained because the patient was unwilling to undergo a skin biopsy.

Discussion

APL is an autosomal recessive genodermatosis reported to be more prevalent in a minority gypsy population within Ireland, known as the Irish travelers[3]. The exact molecular pathogenesis of this condition is unknown. It is hypothesized that the HR gene encodes a transcription co-repressor factor protein that plays an important role in regulating catagen remodeling in the hair cycle. Mutations in this gene cause hair matrix cells to undergo premature and massive apoptosis and separate from the overlying epithelial sheath. Consequently, hair bulbs and dermal papillae become trapped in the dermis, leading to the cessation of hair growth and the formation of keratinous follicular cysts[4,5].

Although originally termed "congenital atrichia" by Ahmad et al.[3], these patients are usually born with normal hair, which is lost permanently within a year of birth. Another distinct feature of APL, as the name suggests, is the development of papular lesions that represent keratin-filled cysts. The lesions are usually generalized, as in our patient, although isolated involvement of the scalp has also been reported by previous authors[5,6]. On trichoscopy, lesions show pinpoint white dots representing cicatricial alopecia, but there are no signs of inflammation, perifollicular pigmentation, or follicular occlusion[6].

APL should be differentiated from alopecia universalis, which also presents with generalized non-scarring hair loss but lacks characteristic papular lesions. Ectodermal dysplasia may be excluded if other ectodermal components, such as teeth, nails, and sweat glands, are normal. VDDR type IIA often presents with congenital atrichia but can be ruled out in the presence of normal serum vitamin D3 and calcium

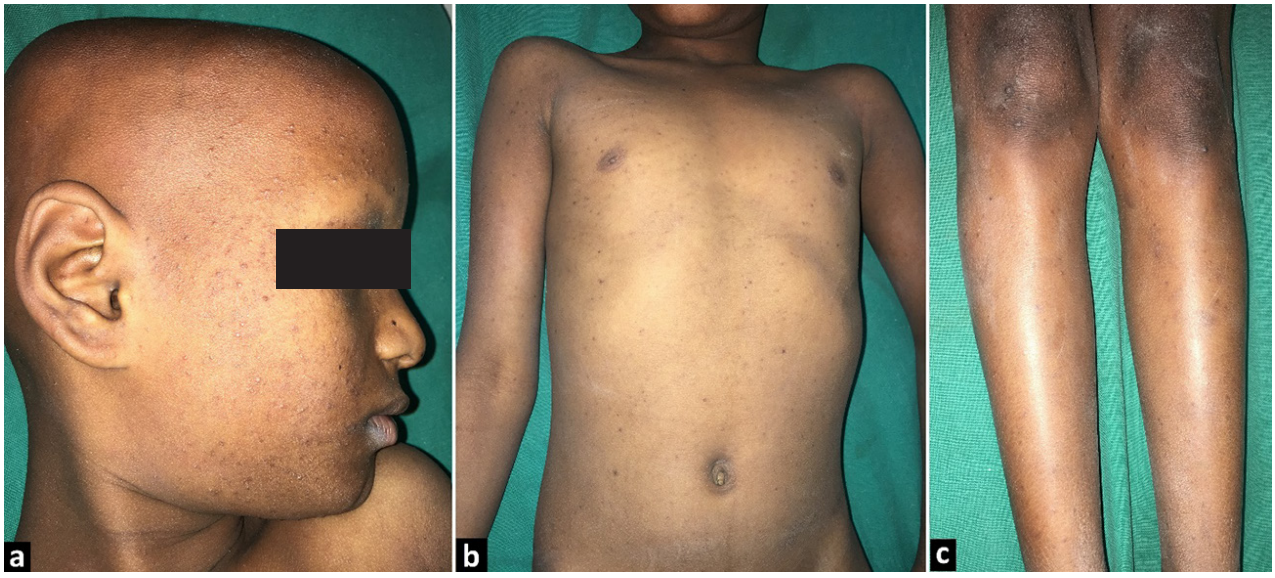

Figure 2. Discrete skin-colored to hyperpigmented papules distributed over the (**a**) scalp and face, (**b**) trunk and arms, and (**c**) legs

Table 1. Revised diagnostic criteria for APL[8]

Major criteria: (4 out of 5 required for diagnosis)
• Permanent and complete disappearance of scalp hair by few months of age. • Few or widespread smooth, whitish or skin-colored milia-like papules on the scalp, face, trunk and extremities from infancy or childhood. • Scalp histology showing replacement of mature hair follicles by follicular cysts filled with cornified material. • Mutation(s) in the human hairless gene detected through genetic testing. • Clinical and/or biochemical exclusion of vitamin-D-dependent rickets
Minor criteria: (supplementary criteria)
• Family history of consanguinity. • Absence of secondary axillary, pubic, or body hair and/or sparse eyebrows and eyelashes. • Normal growth and development, and normal bones, teeth, nails, and sweating. • Whitish hypopigmented streaks on the scalp. • Lack of response to any treatment modality.

levels and normal wrist joint radiographs, as seen in our case. Zlotogorski et al.[7] proposed two main laboratory findings for confirmation of APL diagnosis: i) histopathological evidence of replacement of the lower portions of hair follicles by keratin-filled cysts and ii) molecular evidence of mutations in the HR gene. Yip et al.[8] later published a set of revised diagnostic criteria, as listed in Table 1. Our patient met three major and four minor criteria. Various other conditions have been linked with APL, such as Moynahan syndrome (epilepsy, mental retardation), situs inversus, mesocardia, hidrotic ectodermal dysplasia and premature ageing syndromes[4,6,9]. However, no such association was observed in the present study.

The management of APL is difficult because of the lack of effective treatment options for this condition. Wigs and hair prostheses may be useful camouflage measures. In addition, the psychosocial impact of congenital alopecia is profound and should be adequately addressed in these patients, especially during their formative years[9].

In conclusion, APL is a rare genetic disorder that should be considered when evaluating a child with atrichia. It is important to rule out other conditions with a similar presentation to avoid unnecessary administration of medications and counsel the parents regarding its prognosis.

Ethics

Informed Consent: Written informed consent was obtained from the patient.

Footnotes

Authorship Contributions

Surgical and Medical Practices: B.K.K., A.D., N.R.N.G., S.C., Concept: B.K.K., N.R.N.G., S.C., Design: B.K.K., A.D., N.R.N.G., S.C., Data Collection or Processing: B.K.K., A.D., S.C., Analysis or Interpretation: B.K.K., A.D., N.R.N.G., Literature Search: B.K.K., A.D., N.R.N.G., S.C., Writing: B.K.K., A.D., N.R.N.G., S.C.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Nöthen MM, Cichon S, Vogt IR, et al: A gene for universal congenital alopecia maps to chromosome 8p21-22. Am J Hum Genet. 1998;62:386-90.
2. Sprecher E: Inherited hair disorders. Rook's Textbook of Dermatology. Ed. Griffiths C, Barker J, Bleiker T, Chalmers R, Creamer D. 9th Edition. West Sussex, John Wiley & Sons Ltd., 2016;68:1-24.
3. Ahmad W, Panteleyev AA, Christiano AM: The molecular basis of congenital atrichia in humans and mice: Mutations in the hairless gene. J Investig Dermatol Symp Proc. 1999;4:240-3.
4. Arif T, Adil M, Amin SS: Congenital atrichia with papular lesions: a rare cause of irreversible childhood alopecia. Acta Dermatovenerol Alp Pannonica Adriat. 2018;27:35-6.
5. Thomas M, Daniel S: Atrichia congenita with papular lesions. Indian J Dermatol Venereol Leprol. 2011;77:70-2.
6. Hajare SA, Gavali S, Mukhi J, Singh RP: Congenital atrichia with papular lesions. Dermatol Online J. 2020;26:13030/qt5631x9sh.
7. Zlotogorski A, Panteleyev AA, Aita VM, Christiano AM. Clinical and molecular diagnostic criteria of congenital atrichia with papular lesions. J Invest Dermatol. 2002;118:887-90.
8. Yip L, Horev L, Sinclair R, Zlotogorski A: Atrichia with papular lesions: a report of three novel human hairless gene mutations and a revision of diagnostic criteria. Acta Derm Venereol 2008;88:346-9.
9. Tunuguntla MN, Mathew B, Quadri A, et al: Inherited Hairlessness: A Case Study of Familial Congenital Atrichia. Cureus. 2023;15:e37608.

Case Report
Olgu Sunumu

DOI: 10.4274/turkderm.galenos.2025.38093
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):63-5

Navigating diagnostic challenges in Xeroderma Pigmentosum variant type

Kseroderma Pigmentozum varyant tipinde tanısal zorluklara bakış

Gonca Saraç*, Özlem Dicle*, Yasemin Alanay**, Cuyan Demirkesen***, Sedef Şahin****

*Acıbadem Maslak Hospital, Clinic of Dermatology, İstanbul, Türkiye
**Acıbadem University Faculty of Medicine, Division of Pediatric Genetics, Department of Pediatrics, İstanbul, Türkiye
***Acıbadem University Faculty of Medicine, Department of Pathology, İstanbul, Türkiye
****Acıbadem University Faculty of Medicine, Department of Dermatology, İstanbul, Türkiye

Abstract

Xeroderma pigmentosum (XP) is a rare autosomal recessive genodermatosis caused by mutations in the DNA repair system, leading to impaired repair of ultraviolet (UV) radiation-induced damage. XP is classified into seven nucleotide excision repair-deficient types (XPA to XPG) and a variant type (XPV). Diagnosis can be made at a later age in the XPV subtype, where sunburn reactions are known to be less severe. In this case, a 33-year-old male patient with a history of freckling that began at age 10 and basal cell carcinoma and squamous cell carcinoma in the head and neck region over the past 5 years presented with a suspicious non-pigmented 6 mm nodular lesion in the left subauricular region. Pathological examination revealed a diagnosis of malignant melanoma (MM). Concurrent genetic analysis revealed a homozygous c.491-6T>G mutation in the POLH gene, confirming a diagnosis of XPV. The mild clinical features of XP in our patient made the XPV diagnosis challenging, and the atypical dermoscopic features of the lesion complicated the clinical diagnosis of MM. It is reported that the age of onset of malignant skin tumors in XPV patients is later than in other groups, and the frequency of MM is higher. This case highlights the frequent delay in diagnosis and the diagnostic challenges of skin tumors in XPV patients.
Keywords: Xeroderma pigmentosum, malignant melanoma, dermatoscopy

Öz

Kseroderma pigmentozum (XP), DNA onarım sistemindeki mutasyonlar nedeniyle ultraviyole ışınlarına bağlı hasarın tamirinin bozulduğu nadir bir otozomal resesif genodermatozdur. XP, yedi nükleotid eksizyon onarım eksikliği tipi (XPA'dan XPG'ye) ve bir varyant tip (XPV) olarak sınıflandırılır. Güneş yanığı reaksiyonunun daha az olduğu bilinen XPV alt tipinde tanı daha geç yaşta konulabilmektedir. Sunulmakta olan 10 yaşında başlayan çillenme, son 5 yıldır baş ve boyun bölgesinde ortaya çıkan bazal hücreli karsinom ve skuamöz hücreli karsinom öyküleri olan 33 yaşındaki erkek olguda sol subaurikular bölgede pigmente olmayan 6 mm çapında şüpheli nodüler lezyon tespit edilmiş, patolojik inceleme sonucunda malign melanom (MM) tanısı konulmuştur. Beraberinde yapılan genetik incelemede POLH geninde homozigot c.491-6T>G mutasyonu tespit edilmiş ve hastaya XPV tanısı da konulmuştur. Olgumuzun XP'ye ait hafif klinik bulguları XPV tanısını, lezyonun atipik dermatoskopik özellikleri de klinik olarak MM tanısını zorlu kılmıştır. XPV hastalarında malign deri tümörlerinin görülme yaşı diğer gruplara göre daha geç, MM görülme sıklığının ise daha yüksek olduğu bildirilmektedir. Bu olgu, XPV hastalarında tanının gecikme sıklığını ve deri tümörlerinin tanısal zorluklarını vurgulamaktadır.
Anahtar Kelimeler: Kseroderma pigmentozum, malign melanom, dermatoskopi

Address for Correspondence/Yazışma Adresi: Gonca Saraç MD, Acıbadem Healthcare Group, Maslak Hospital, İstanbul, Türkiye
E-mail: goncasarac@yahoo.com **ORCID:** orcid.org/0000-0002-1263-6394
Received/Geliş Tarihi: 07.08.2024 **Accepted/Kabul Tarihi:** 19.05.2025 **Publication Date/Yayınlanma Tarihi:** 01.07.2025

Cite this article as/Atıf: Saraç G, Dicle Ö, Alanay Y, Demirkesen C, Şahin S. Navigating diagnostic challenges in Xeroderma Pigmentosum variant type. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):63-5

Introduction

Xeroderma pigmentosum (XP) is a rare autosomal recessive genodermatosis caused by mutations in the DNA repair system, leading to impaired repair of UV radiation-induced damage. This deficiency results in early-onset actinic damage and the development of skin tumors. XP is classified into seven nucleotide excision repair-deficient types (Xeroderma pigmentosum complementation group A through Xeroderma Pigmentosum, complementation group G) and a xeroderma pigmentosum variant type (XPV). The manifestation of sunburn reactions and actinic damage varies according to the complementation group, affecting the timing of diagnosis and the frequency of skin cancer development based on cumulative UV radiation exposure[1]. Nevertheless, differentiating malignant lesions in the actinically damaged skin of all these patients is challenging. Although dermoscopy is indispensable in this context, it requires greater attention, and the threshold for biopsy should be low.

Case Report

A 33-year-old male patient, with no skin findings at birth, reported a history of freckling around the age of 10. In his twenties, he experienced febrile convulsions, requiring medication for 4-5 years. Consanguinity was noted in his parental lineage. The patient exhibited actinic damage inconsistent with his age, numerous seborrheic keratoses, and fibroepithelial polyps, and from the age of 27, he began developing non-melanoma skin tumors. Over five years, during his sporadic visits to the dermatology clinic, he underwent multiple excisions of basal cell carcinoma (BCC) and squamous cell carcinoma from the head and neck region. Despite occupational sun exposure, the patient exhibited inadequate adherence to sun protection practices. Comprehensive evaluations revealed no accompanying ophthalmological, neurological, or cardiological pathology. During follow-up, a newly developed erythematous, centrally crusted nodular lesion measuring 6 mm in diameter was identified in the left subauricular region of the patient's neck (Figure 1). Dermoscopic examination revealed a non-pigmented, irregularly bordered neoplasm featuring linear-irregular and hairpin vessels along the periphery and a central yellow crust (Figure 2). A punch biopsy was performed under the presumption of keratoacanthoma or squamous cell carcinoma; however, pathological examination revealed malignant melanoma (MM) (Figure 3). Subsequent excision and sentinel lymph node examination confirmed nodular MM with a Breslow thickness of 3.4 mm (pT3b) and sentinel lymph node involvement. No distant metastases were identified.

Genetic consultation and whole-exome analysis revealed a homozygous c.491-6T>G mutation in the *POLH* gene, confirming the patient's XPV diagnosis at the age of 33.

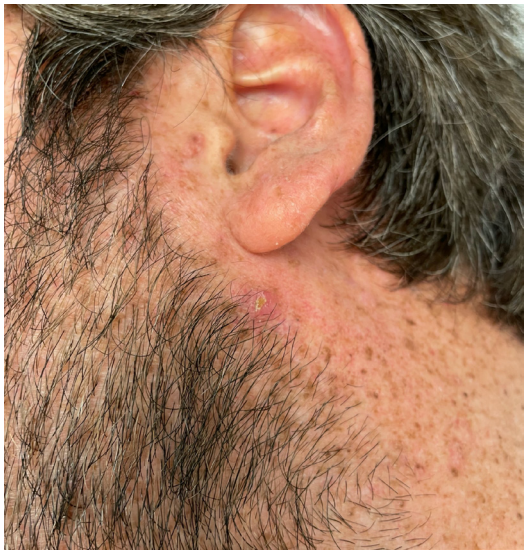

Figure 1. Clinical presentation of subauricular nodular lesion

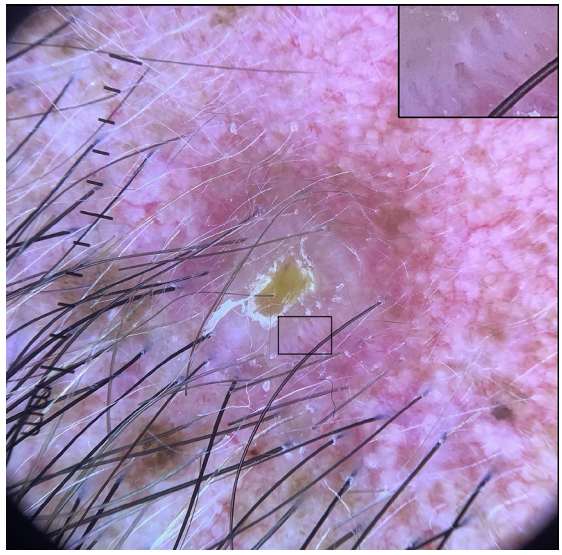

Figure 2. Dermoscopy of the lesion, showing a non-pigmented nodule with linear-irregular and hairpin vessels (square, enlarged)

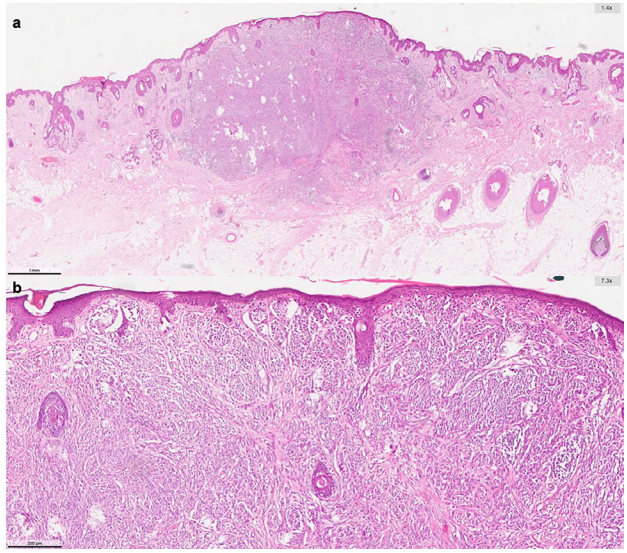

Figure 3. Histopathological features of the malignant melanoma lesion. A spindle cell tumor occupying the entire dermis and presenting with an ulcerated surface is observed. **(a)** Hematoxylin and eosin, x2; **(b)** Hematoxylin and eosin, x10

Discussion

XP subtypes A-G result from defects in the nucleotide excision repair (NER) pathway, rendering the removal of UV-induced photoproducts defective. XPV is characterized by a deficiency in post-replication repair rather than NER, primarily due to the *POLH* gene defect. This gene is involved in DNA synthesis after damage (translesion synthesis process), and POLH mutations in XPV patients result in a decreased ability of postreplication repair of damaged DNA following UV exposure, leading to the mutagenic effects of UV and subsequent skin cancers[2]. Unlike other groups, genetic complementation tests are not used for XPV diagnosis; instead, *POLH* gene examination is performed[3].

In the classic XP phenotype, freckling appears before the age of 2, and severe sunburns occur with minimal sun exposure. The median age for the first non-melanoma skin cancer is reported to be 9, and for melanoma, it is 22[4]. XPV typically presents with less severe clinical manifestations of UV sensitivity compared to other groups. Although abnormal pigmentation responses such as freckling and lentigines occur in these patients, they have normal minimal erythema dose values, and sunburn reactions are not as severe, with some even retaining the ability to tan[5,6]. Consequently, sun protection behaviors in XPV patients are not as early and stringent as in other groups, leading to higher skin cancer prevalence due to delayed diagnosis and increased sun exposure[7]. XPV patients generally do not exhibit neurodegeneration seen in other types, and long-term survival is better[4].

XPV accounts for approximately 25% of all XP cases. In Japan, 80% of XPV patients have a history of skin cancers, including BCC in 63%, squamous cell carcinoma in 30.4%, and MM in 23.9% of cases[7]. Skin cancers in this group appear at a later age compared to classic XP patients, with the first BCC occurring at an average age of 41.5[6]. Notably, MM is significantly more common in XPV patients than in other groups[7].

Identifying and clinically distinguishing skin tumors in XP patients, who have severe underlying actinic damage, is more challenging than in normal individuals. Dermoscopy remains an indispensable diagnostic tool, with the features sought in XP patients' skin tumors being similar to those in normal individuals. In XP patients, MM lesions most commonly present with asymmetry, multiple colors, prominent pigment networks, blue-gray areas, and atypical globules/dots[8,9].

The MM lesion in our patient lacked a pigment network; the vascular pattern within such lesions may aid in malignancy diagnosis. Although linear-irregular vessels suggestive of melanoma are observed, the presence of hairpin vessels with occasional peripheral halo and crusting in dermoscopy is a characteristic more commonly associated with keratinizing tumors. Nonetheless, the observation of this dermoscopic pattern in an XP patient strongly raises suspicion of a malignant lesion. This case highlights the frequency of delayed diagnosis in XPV patients and the challenges in diagnosing skin tumors in these patients. It also underscores the difficulty in diagnosing skin tumors, especially amelanotic melanoma lesions, even dermoscopically in actinically damaged skin. Due to the higher incidence of melanoma and overall increased frequency of skin tumors, dermatologists should consider the variant subtype of XP in patients with abnormal pigmentation changes but no rapid and severe sunburn history.

Ethics

Informed Consent: The patient in this manuscript has given written informed consent to the publication of their case details and images.

Footnotes

Authorship Contributions

Surgical and Medical Practices: G.S., Ö.D., Y.A., C.D., S.Ş., Literature Search: G.S., Writing: G.S., Ö.D., S.Ş.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Lehmann AR, McGibbon D, Stefanini M. Xeroderma pigmentosum. Orphanet J Rare Dis. 2011;6:70.
2. Gratchev A, Strein P, Utikal J, Sergij G. Molecular genetics of Xeroderma pigmentosum variant. Exp Dermatol. 2003;12:529-36.
3. Tanioka M, Masaki T, Ono R, et al.: Molecular analysis of DNA polymerase eta gene in Japanese patients diagnosed as xeroderma pigmentosum variant type. J Invest Dermatol. 2007;127:1745-51.
4. Black JO: Xeroderma Pigmentosum. Head Neck Pathol. 2016; 10: 139-44.
5. Leung AK, Barankin B, Lam JM, Leong KF, Hon KL: Xeroderma pigmentosum: an updated review. Drugs Context. 2022;11:2022-2-5.
6. Moriwaki S, Kanda F, Hayashi M, et al.: Xeroderma pigmentosum clinical practice guidelines. J Dermatol. 2017;44:1087-96.
7. Nishigori C, Nakano E, Masaki T, et al.: Characteristics of Xeroderma Pigmentosum in Japan: Lessons From Two Clinical Surveys and Measures for Patient Care. Photochem Photobiol. 2019;95:140-53.
8. Malvehy J, Puig S, Martí-Laborda RM: Dermoscopy of skin lesions in two patients with xeroderma pigmentosum. Br J Dermatol. 2005;152:271-8.
9. Faria Licarião Rocha LK, Ferreira P, Avancini J, et al.: Dermoscopic features of 61 skin lesions in xeroderma pigmentosum patients: A cross-sectional study. J Am Acad Dermatol. 2022;86:1361-2.

Case Report
Olgu Sunumu

DOI: 10.4274/turkderm.galenos.2025.63373
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):66-9

A new *HLA* susceptibility haplotype defined in three familial cases of frontal fibrosing alopecia

Üç ailevi frontal fibrozan alopesi vakasında tanımlanan yeni bir HLA duyarlılık haplotipi

Ahmet Kağan Özdemir*, Yıldız Hayran**, Füsun Özmen***, Melek Günindi Korkut***, Seray Külcü Çakmak**

*Erzincan Binali Yıldırım University Faculty of Medicine, Department of Dermatology, Erzincan, Türkiye
**Ankara Bilkent City Hospital, Clinic of Dermatology, Ankara, Türkiye
***Hacettepe University Oncology Hospital, Clinic of Basic Oncology, Ankara, Türkiye

Abstract

Frontal fibrosing alopecia (FFA) is a scarring alopecia that primarily affects postmenopausal women. Although its etiology remains unknown, familial cases suggest a genetic basis. In this case report, we present the findings of three sisters with familial FFA, aged 55, 60, and 62 years, from Türkiye. Genetic analysis revealed that all three sisters shared the *human leukocyte antigen (HLA)-A*11:01; B*35:01; C*04:01* haplotypes. Two sisters had *HLA-DRB1*03:01* and *HLA-DQB1*02:01*, while the third had *HLA-DRB1*01:01; HLA-DQB1*05:01*. The shared *HLA-A*11:01*, *B*35:01*, and *C*04:01* haplotype has not been previously associated with familial FFA. This finding marks the first familial FFA report from Türkiye and suggests a new genetic susceptibility haplotype for FFA in the Turkish population. The variation in *HLA-DRB1* and *HLA-DQB1* alleles among the sisters indicates complex genetic influences on the familial FFA. Further research is required to determine the role of these genetic variations in disease progression and to identify potential therapeutic approaches.
Keywords: Familial frontal fibrosing alopecia, *HLA* haplotypes, genetic susceptibility, scarring alopecia

Öz

Frontal fibrozan alopesi (FFA), genellikle menopoz sonrası kadınları etkileyen skatrisyel bir alopesidir. Etiyolojisi bilinmemekle birlikte, ailesel olgular genetik bir bileşen öne sürmektedir. Bu olgu raporunda, Türkiye'den 55, 60 ve 62 yaşlarındaki üç kız kardeşle ilgili ailesel FFA bulgularını paylaşıyoruz. Genetik analiz, üç kardeşin de insan lökosit antijeni *(HLA)-A*11:01; B*35:01; C*04:01* haplotipini paylaştığını ortaya koydu. İki kardeş *HLA-DRB1*03:01; HLA-DQB1*02:01*, diğer kardeş ise *HLA-DRB1*01:01; HLA-DQB1*05:01* alellerine sahipti. Paylaşılan *HLA-A*11:01; B*35:01; C*04:01* haplotipi, literatürde daha önce ailesel FFA ile ilişkilendirilmemiştir. Bu bulgular, Türkiye'den bildirilen ilk ailesel FFA olgusunu oluşturmaktadır ve FFA için yeni bir genetik yatkınlık haplotipini öne sürmektedir. Kız kardeşler arasındaki *HLA-DRB1* ve *HLA-DQB1* alel varyasyonları, ailesel FFA üzerindeki karmaşık genetik etkileri vurgulamaktadır. Bu genetik varyasyonların hastalık progresyonu ve potansiyel terapötik yaklaşımlar üzerindeki rolünü belirlemek için daha fazla araştırmaya ihtiyaç vardır.
Anahtar Kelimeler: Ailesel frontal fibrozan alopesi, *HLA* haplotipleri, genetik yatkınlık, skatrisyel alopesi

Address for Correspondence/Yazışma Adresi: Ahmet Kağan Özdemir MD, Erzincan Binali Yıldırım University Faculty of Medicine, Department of Dermatology, Erzincan, Türkiye
E-mail: ahmetkaganozdemir@gmail.com **ORCID:** orcid.org/0000-0002-0930-7215
Received/Geliş Tarihi: 12.09.2024 **Accepted/Kabul Tarihi:** 23.05.2025 **Publication Date/Yayınlanma Tarihi:** 01.07.2025

Cite this article as/Atıf: Özdemir AK, Hayran Y, Özmen F, Günindi Korkut M, Külcü Çakmak S. A new HLA susceptibility haplotype defined in three familial cases of frontal fibrosing alopecia. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):66-9

Introduction

Frontal fibrosing alopecia (FFA) belongs to the group of primary cicatricial alopecias, most commonly presents as progressive recession of the frontotemporal hairline accompanied by bilateral eyebrow loss, particularly among postmenopausal women[1,2]. Since its initial description by Kossard[3] in 1994, there has been a marked global rise in FFA incidence, and it is now recognized as the most frequently reported form of cicatricial alopecia in numerous studies[3,4].

The precise pathogenesis of FFA remains to be elucidated; however, multiple factors, such as hormonal changes, immune system dysregulation, and environmental influences have been proposed[1]. Although sporadic cases are the norm, the presence of familial clusters suggests an underlying genetic and/or epigenetic component[5-12]. A positive family history has been reported in as many as 8% of cases, raising the possibility of an autosomal dominant mode of inheritance with incomplete penetrance[2,13].

FFA is considered a clinical subtype of lichen planopilaris. Although the two conditions display distinct clinical features, they both fall under the category of lymphocytic primary scarring alopecias and share similar immune and inflammatory mechanisms[14]. The disease process likely begins with the collapse of immune privilege at the level of the hair follicle (HF) bulge, where epithelial HF stem cells (eHFSCs) reside. This disruption enables T cell-mediated inflammatory responses, resulting in eHFSC apoptosis and subsequent irreversible alopecia[15]. The immune privilege of HF protects eHFSCs from autoimmune attacks by suppressing the expression of major histocompatibility complex (MHC) class I and II molecules[16,17]. Aberrant expression of *human leukocyte antigen (HLA)* class I and II molecules, which are components of MHC, within eHFSCs may compromise this immune privilege[15]. While *HLA* class I molecules (A, B, and C) are present on all nucleated cells, *HLA* class II molecules (such as *DRB1, DQB1,* and *DPB1*) are typically expressed on immune-activated cells. Variants within *HLA* genes have been implicated in altering susceptibility to a wide range of infectious, inflammatory, and autoimmune diseases[18].

Few studies have reported shared *HLA* haplotypes in familial FFA cases. To date, three distinct susceptibility haplotypes have been documented across different patient cohorts[19-21]. Furthermore, *HLA* class II polymorphisms previously linked to conditions such as Lassueur-Graham-Little-Piccardi syndrome and both familial and sporadic forms of lichen planus were not associated with familial FFA[9,22-25].

In this report, we present a newly identified *HLA* susceptibility haplotype in three sisters affected by familial FFA, representing the first such case reported in Türkiye. Our findings contribute to the growing body of evidence that genetic predisposition plays a pivotal role in FFA pathogenesis. By analyzing the genetic profiles of affected individuals, we aimed to enhance our understanding of the hereditary factors involved in this disease.

Case Report

Three sisters, aged 62, 60, and 55 years, presented with progressive frontotemporal hair loss and lateral eyebrow alopecia (Figures 1, 2, and 3). All patients' symptoms began after menopause and gradually worsened. None of the patients experienced body hair loss or facial pain. They had no other siblings. Their personal and family medical histories were unremarkable, with no evidence of dermatologic or autoimmune conditions. They lived in the same city but resided in different neighborhoods. They did not work in regular jobs and mostly stayed at home. They did not regularly use cosmetic products, such as sunscreen. None of the patients were smokers. Trichoscopy revealed loss of hair follicular openings in the affected hairline and perifollicular hyperkeratosis in all patients (Figure 4). Histopathological examination was compatible with FFA. We addressed the *HLA-A, HLA-B, HLA-C, HLA-DRB1, and HLA-DQB1* genetic variabilities in this family. Haplotype analysis was performed using the sequence-specific oligonucleotide method. Haplotype analysis revealed *HLA-A*11:01; B*35:01; C*04:01* shared among all sisters. Two sisters had *HLA-DRB1*03:01 and HLA-DQB1*02:01*, while the other sister had *HLA-DRB1*01:01; HLA-DQB1*05:01*. We were unable to do genetic analysis on the patient's mother and father since they were deceased, and on their offspring because they were unable to attend the hospital.

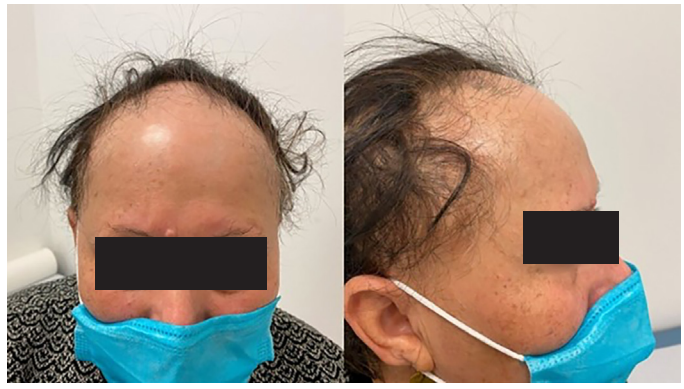

Figure 1. Oldest sister

Figure 2. Middle sister

Figure 3. Youngest sister

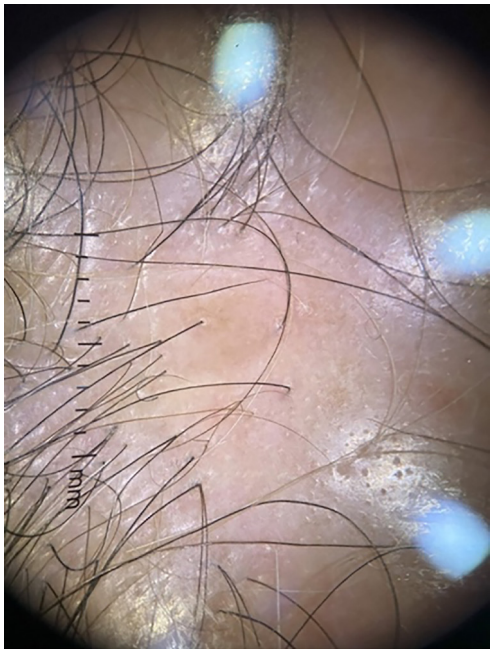

Figure 4. Trichoscopic examination showing loss of hair follicular openings in the affected hairline and perifollicular hyperkeratosis

Discussion

In this family with FFA, a shared *HLA-A*11:01, B*35:01*, and *C*04:01* haplotype was identified across all three sisters. To date, this specific HLA class I haplotype has not been linked to FFA in the literature. Furthermore, this report is the first to identify familial FFA in the Turkish population.

Earlier studies have highlighted several other HLA haplotypes that are potentially associated with FFA. A genome-wide association study by Tziotzios et al.[19] conducted in British and Spanish cohorts revealed four genomic regions implicated in FFA, one of which included the *HLA-B*07:02* allele. Additionally, this study highlighted the associations of genetic variants involved in xenobiotic metabolism, T-cell regulation, and antigen presentation across four susceptibility loci. The *HLA-B*07:02* allele demonstrated the strongest single-locus association, conferring an approximate four-fold increase in FFA risk among women. Rayinda et al.[26] subsequently confirmed the significant contribution of this allele to FFA risk in men. Despite the limited number of familial cases included, these findings from large cohort studies underscore the role of genetic predisposition in FFA.

Porriño-Bustamante et al.[21] described an alternate susceptibility haplotype- *HLA-A*33:01; B*14:02; C*08:02*-in 13 Spanish familial FFA cases. The study also reported a link between the *HLA* class I haplotype *F16A* and the *CYP21A2 p.V281L* mutation in affected individuals, suggesting that this mutation, previously associated with congenital adrenal hyperplasia, may serve as a genetic marker for familial FFA.

Additionally, Ramos et al.[20] analyzed a Brazilian cohort of both familial and sporadic FFA cases and identified two potential susceptibility haplotypes: *HLA-B*07:02:01:01; C*07:02:01:03* and *HLA-B*42:01:01:01; C*17:01:01:02*[20]. The first haplotype had not been previously associated with FFA, and both were found in certain unaffected relatives, implying that environmental or epigenetic factors may influence disease expression.

Conclusion

Our identification of the *HLA-A*11:01, B*35:01,* and *C*04:01* haplotypes in this Turkish family provides further evidence supporting the genetic contribution to FFA pathogenesis. While prior studies have described various *HLA* haplotypes associated with FFA, our findings suggest possible ethnic and regional differences in genetic susceptibility. The variation in *HLA-DRB1* and *HLA-DQB1* alleles observed among the siblings further underscores the complex genetic architecture underlying the expression of this disease. Familial cases such as this one offer valuable insights into the genetic mechanisms driving FFA. Ongoing research is required to elucidate how these genetic factors interact with additional risk elements to inform the development of targeted therapeutic strategies.

Ethics

Informed Consent: Written consent was obtained from all patients for the use of their images and genetic material.

Footnotes

Authorship Contributions

Surgical and Medical Practices: A.K.Ö., Y.H., F.Ö., M.G.K., S.K.Ç., Concept: A.K.Ö., Y.H., F.Ö., M.G.K., S.K.Ç., Design: A.K.Ö., Y.H., F.Ö., M.G.K., S.K.Ç., Data Collection or Processing: A.K.Ö.,Y.H., F.Ö., M.G.K., S.K.Ç., Analysis or Interpretation: A.K.Ö., Y.H., F.Ö., M.G.K., S.K.Ç., Literature Search: A.K.Ö., Y.H., F.Ö., M.G.K., S.K.Ç., Writing: A.K.Ö., Y.H., F.Ö., M.G.K., S.K.Ç.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Mirmirani P, Tosti A, Goldberg L, Whiting D, Sotoodian B: Frontal frontal fibrosing alopecia: An emerging epidemic. Skin Appendage Disord. 2019;5:90-3.
2. Kerkemeyer KLS, Eisman S, Bhoyrul B, Pinczewski J, Sinclair RD: Frontal fibrosing alopecia. Clin Dermatol. 2021;39:183-93.
3. Kossard S: Postmenopausal frontal fibrosing alopecia. Scarring alopecia in a pattern distribution. Arch Dermatol. 1994;130:770-4.
4. Vañó-Galván S, Saceda-Corralo D, Blume-Peytavi U, et al: Frequency of the types of alopecia at twenty-two specialist hair clinics: A multicenter study. Skin Appendage Disord. 2019;5:309-15.
5. Photiou L, Nixon RL, Tam M, Green J, Yip L: An update of the pathogenesis of frontal fibrosing alopecia: What does the current evidence tell us? Australas J. 2019;60:99-104.
6. Junqueira Ribeiro Pereira AF, Vincenzi C, Tosti A: Frontal fibrosing alopecia in two sisters. Br J Dermatol. 2010;162:1154-5.
7. Tziotzios C, Fenton DA, Stefanato CM, McGrath JA: Familial frontal fibrosing alopecia. J Am Acad Dermatol. 2015;73:e37.
8. Dlova N, Goh CL, Tosti A: Familial frontal fibrosing alopecia. Br J Dermatol. 2013;168:220-2.
9. Rivas MM, Antolín SC, Sambucety PS, González ES, Ruíz de Morales JM, Prieto MÁ: Frontal fibrosing alopecia and lichen planopilaris in HLA-identical mother and daughter. Indian J Dermatol Venereol Leprol. 2015;81:162-5.
10. Navarro-Belmonte MR, Navarro-López V, Ramírez-Boscà A, et al: Case series of familial frontal fibrosing alopecia and a review of the literature. J Cosmet Dermatol. 2015;14:64-9.

11. Starek JV, Raszl TP, Kaddourah SMEH: Frontal fibrosing alopecia: report of four sisters. An Bras Dermatol. 2023;98:855-8.
12. Carpanese MA, Guglielmo A, Alessandrini A, Bruni F, Piraccini B, Starace M: Familial frontal fibrosing alopecia: the first Italian case. Ital J Dermatol Venerol. 2022;157:457-8.
13. Vañó-Galván S, Molina-Ruiz AM, Serrano-Falcón C, et al: Frontal fibrosing alopecia: a multicenter review of 355 patients. J Am Acad Dermatol. 2014;70:670-8.
14. Miao YJ, Jing J, Du XF, Mao MQ, Yang XS, Lv ZF: Frontal fibrosing alopecia: A review of disease pathogenesis. Front Med (Lausanne). 2022;9:911944.
15. Harries MJ, Meyer K, Chaudhry I, et al: Lichen planopilaris is characterized by immune privilege collapse of the hair follicle's epithelial stem cell niche. J Pathol. 2013;231:236-47.
16. Ito T, Ito N, Saatoff M, et al: Maintenance of hair follicle immune privilege is linked to prevention of NK cell attack. J Invest Dermatol. 2008;128:1196-206.
17. Christoph T, Müller-Röver S, Audring H, et al: The human hair follicle immune system: cellular composition and immune privilege. Br J Dermatol. 2000;142:862-73.
18. Fernando MM, Stevens CR, Walsh EC, et al: Defining the role of the MHC in autoimmunity: a review and pooled analysis. PLoS Genet. 2008;4:e1000024.
19. Tziotzios C, Petridis C, Dand N, et al: Genome-wide association study in frontal fibrosing alopecia identifies four susceptibility loci including HLA-B*07:02. Nat Commun. 2019;10:1150.
20. Ramos PM, Garbers L, Silva NSB, et al: A large familial cluster and sporadic cases of frontal fibrosing alopecia in Brazil reinforce known human leucocyte antigen (HLA) associations and indicate new HLA susceptibility haplotypes. J Eur Acad Dermatol Venereol 2020;34:2409-13.
21. Porriño-Bustamante ML, López-Nevot MÁ, Aneiros-Fernández J, et al: Study of Human Leukocyte Antigen (HLA) in 13 cases of familial frontal fibrosing alopecia: CYP21A2 gene p.V281L mutation from congenital adrenal hyperplasia linked to HLA class I haplotype HLA-A*33:01; B*14:02; C*08:02 as a genetic marker. Australas J Dermatol. 2019;60:195-200.
22. Powell FC, Rogers RS, Dickson ER, Moore SB: An association between HLA DRI and lichen planus. Br J Dermatol. 1986;114:473-8.
23. Katzenelson V, Lotem M, Sandbank M: Familial lichen planus. Dermatologica. 1990;180:166-8.
24. Viglizzo G, Verrini A, Rongioletti F: Familial Lassueur-Graham-Little-Piccardi syndrome. Dermatology. 2004;208:142-4.
25. Chan DV, Kartono F, Ziegler R, et al: Absence of HLA-DR1 positivity in 2 familial cases of frontal fibrosing alopecia. J Am Acad Dermatol. 2014;71:208-10.
26. Rayinda T, McSweeney SM, Fenton D, et al: Shared genetic risk variants in both male and female frontal fibrosing alopecia. J Invest Dermatol. 2023;143:2311-4.

Letter to the Editor
Editöre Mektup

DOI: 10.4274/turkderm.galenos.2020.25348
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):70-1

A facial nodular orf infection in an HIV positive patient: Report of a case

HIV pozitif hastada yüzde nodüler orf enfeksiyonu: Olgu sunumu

Sema Aytekin*, Burak Ekinci, Cansu Sönmez*****

*Tekirdağ Namık Kemal University Faculty of Medicine, Department of Dermatology, Tekirdağ, Türkiye
**Erbaa Public Hospital, Clinic of Dermatology, Tokat, Türkiye
***Haydarpaşa Numune Training ang Research Hospital, Clinic of Pathology, İstanbul, Türkiye

Keywords: Orf, HIV, face

Anahtar Kelimeler: Orf, HIV, yüz

To Editor,

Orf infection in humans is most commonly characterized by a solitary, nodular lesion affecting the fingers or hands[1-4]. Involvement of the face has rarely been reported. We report a huge orf lesion on the face of a young male.

A 32-year-old human immunodeficiency virus (HIV)-positive man presented with a rapidly growing, painless nodule on his right cheek, characterized by oozing and bleeding. He had first noticed the lesion approximately 2 weeks ago. There was no history of trauma, burn, pre-existing skin lesion, or topical application at the site of the lesion. He had no fever, chills, or systemic symptoms. He was working as a shepherd. Four years ago, he was diagnosed with HIV while being treated at an inpatient clinic due to the sudden onset of severe psoriasis and widespread Molluscum contagiosum lesions. Dermatological examination revealed a noticeably raised red nodule about 3x2 cm in size on the right cheek (Figure 1 a, b.) No palpable lymphadenopathy was detected, and systemic examination revealed no abnormalities. Routine hematological and biochemical tests were within normal ranges. He had been receiving HIV treatment (tenofovir, disoproxil, emtricitabine, and dolutegravir) for approximately two years, and his HIV- ribonucleic acid was negative and his CD4+ T lymphocyte count was 600/mm^3.

The histopathologic examination revealed focal irregular acanthosis, vascular proliferation, diffuse polymorphonuclear leukocyte infiltration, edema, abscess formation, and rupture of follicle epithelium in the dermis (Figure 1c, d). We only used 10% povidone iodine-containing local antiseptic twice daily for two weeks. Significant improvement was observed on the 10th day and regressed during the following 6 weeks without scarring.

Ecthyma contagiosum (Orf) is an uncommon, benign, self-limiting infection resulting from the cutaneous inoculation of a parapoxvirus in humans. This resistant virus may be transmitted to humans either by direct contact with infected animals or through objects or, less commonly through contaminated meat[1-4]. In our country, the orf disease is generally transmitted during sheep slaughter at the feast of sacrifice[2]. Orf typically affects the hands and fingers, with

Address for Correspondence/Yazışma Adresi: Sema Aytekin MD, Tekirdağ Namık Kemal University Faculty of Medicine, Department of Dermatology, Tekirdağ, Türkiye
E-mail: semaaytekin@yahoo.com **ORCID:** orcid.org/0000-0003-1376-1573
Received/Geliş Tarihi: 18.12.2020 **Accepted/Kabul Tarihi:** 24.03.2025 **Publication Date/Yayınlanma Tarihi:** 01.07.2025
Cite this article as/Atıf: Aytekin S, Ekinci B, Sönmez C. A facial nodular orf infection in an HIV positive patient: report of a case. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):70-1

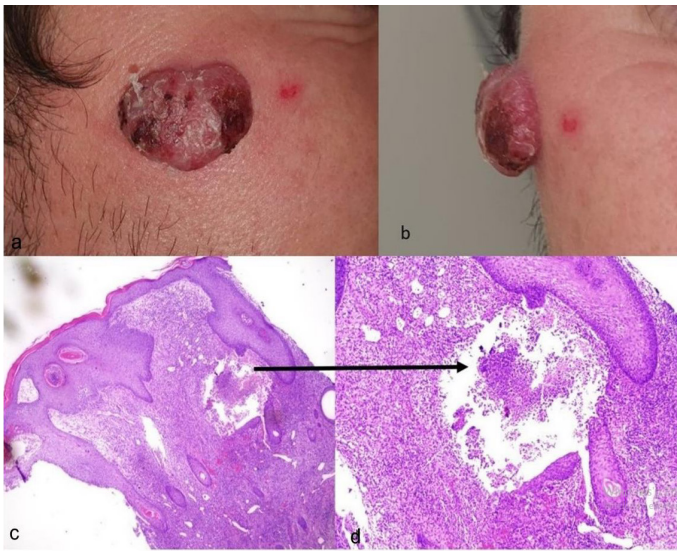

Figure 1. a, b. Oozy and bloody nodule on the right cheek, c. Focal irregular acanthosis, crust in epidermis and capillary vessel proliferation, diffuse polymorph leukocyte infiltration in dermis (HEX40), d. edema, abscess formation, rupture in follicle epithelium in dermis (HEX100).
HE: Hematoxylin and Eosin

facial lesions being a much less common presentation[3]. Giant orf on the nose has been reported in a few cases, one of which was a 14-month-old baby[3-5]. Orf virus causes pyogenic granuloma(PG)-like lesions, which are characterized histopathologically by massive capillary proliferation and dilation[6].

People with significant T-cell dysfunction may develop atypical lesions, such as giant orf and/or have an unusual course of illness. Our patient might have had a giant lesion since HIV+ patients have a high risk of endothelial dysfunction[1,7].

Since the diagnosis of human Orf is largely clinical, physicians must be aware of the typical lesion progression to avoid diagnostic errors or inappropriate interventions. As Orf lesions resolve in about four to six weeks without scarring, biopsy should generally be avoided. If the clinical diagnosis is unclear, confirmation may be obtained through viral culture and electron microscopic examination of lesion aspirates. If these opportunities are not available, a biopsy should be taken.

The actual incidence of orf in Türkiye is unclear, as it is not a reportable disease. Physicians should maintain a high index of suspicion for orf when evaluating localized skin lesions in patients with relevant animal exposure.

Differential diagnosis of orf includes other infectious diseases, PG, and keratoacanthoma[1-3]. Multiple orf lesions, especially developing on burn scars, are misdiagnosed as eruptive or generalized PG in the literature. When viral cytopathic effects are observed in histopathological examination in PG-like lesions, orf should be considered[6].

In healthy patients, a conservative approach should be applied, as most cases spontaneously resolve within 6-8 weeks. Local antiseptics may be used to prevent secondary bacterial infections[8]. We also used povidone-iodine-containing local antiseptic. Significant improvement was observed on the 10th day. However, treatment options for giant or multiple orf lesions in immunocompromised individuals include cryotherapy, topical imiquimod, intralesional cidofovir, and intralesional interferon alfa injections[1,3].

Ethics

Informed Consent: Informed consent was obtained from the patient for the publication of his images and medical information.

Footnotes

Authorship Contributions

Concept: S.A., Design: S.A., Data Collection or Processing: B.E., C.S., Analysis or Interpretation: S.A., B.E., Writing: S.A.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Lederman ER, Green GM, DeGroot HE, et al: Progressive orf virus infection in a patient with lymphoma: Successful treatment using imiquimod. Clin Infect Dis. 2007;44:100-3.
2. Gündüz K, Inanir I, Sacar T: Orf and religious practices. J Dermatol. 2005;32:306-8.
3. Key SJ, Catania J, Mustafa SF, et al: Unusual presentation of human giant orf (ecthyma contagiosum). J Craniofac Surg. 2007;18:1076-8.
4. Ata N, Göğüş HE, Kiliç S: Giant orf on the nose. J Craniofac Surg. 2017;28:234-5.
5. Sleiay M, Alqreea M, Alabdullah H: A 4-month-old male baby with an orf lesion on his nose: a rare case report. Ann Med Surg (Lond). 2023;86:545-7.
6. Biyik Ozkaya D, Taskin B, Tas B, et al: Poxvirus-induced angiogenesis after a thermal burn. J Dermatol. 2014;41:830-3.
7. Mazzuca P, Caruso A, Caccuri F: HIV-1 infection, microenvironment and endothelial cell dysfunction. New Microbiol. 2016;39:163-73.
8. Bigliardi PL, Alsagoff SAL, El-Kafrawi HY, Pyon JK, Wa CTC, Villa MA: Povidone iodine in wound healing: A review of current concepts and practices. Int J Surg. 2017;44:260-8.

Letter to the Editor
Editöre Mektup

DOI: 10.4274/turkderm.galenos.2024.16974
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):72-3

Psoriasis exacerbation following intravesical BCG immunotherapy for bladder carcinoma: A case report

Mesane kanseri tedavisinde intravezikal BCG immünoterapisi kullanımı sonrası psöriazis alevlenmesi: bir olgu sunumu

Yusuf Can Edek, Ahmet Soyugür, Esra Adışen

Gazi University Faculty of Medicine, Department of Dermatology, Ankara, Türkiye

Keywords: BCG, malignancy, psoriasis

Anahtar Kelimeler: BCG, malignite, psöriazis

To Editor,

Psoriasis is a chronic, immune-mediated inflammatory disease with a broad spectrum of clinical manifestations that can be out or exacerbated by genetic and environmental factors[1]. Exacerbations of psoriasis have been observed after administering influenza, tetanus-diphtheria, and Bacillus Calmette-Guérin (BCG) vaccines[2-4]. Additionally, BCG immunotherapy, used to treat bladder cancer, may be one of the causes of psoriasis flare-ups[5]. Here, we present a male patient with psoriasis vulgaris who had psoriasis exacerbation following intravesical BCG immunotherapy for bladder carcinoma treatment.

A 58-year-old male with a history of psoriasis vulgaris applied to our dermatology outpatient clinic due to a flare-up of his lesions. He was diagnosed with psoriasis 23 years ago and used topical treatments, methotrexate, cyclosporine, infliximab, and adalimumab for the disease in the past. Over the past two years, his psoriasis had been completely clear under ustekinumab. After he was diagnosed with invasive urothelial carcinoma, ustekinumab was stopped. Three months after taking intravesical BCG immunotherapy, one week after the third dose, pruritic lesions occurred on his body. He had no recent history of infection or allergy. Dermatologic examination revealed widespread, pruritic erythematous plaques on the trunk and extremities, and pitting on the nails. (Figure 1) Histopathological analysis of a punch biopsy from the plaque on the trunk was compatible with psoriasis. Based on clinical examination and histopathological analysis, the patient was accepted as psoriasis flare-up. When the patient's complaints appeared one week after the third BCG immunotherapy, and the Naranjo Adverse Reaction score calculated was 5 (5-8: possible), psoriasis exacerbation was considered secondary to intravesical BCG immunotherapy. The Psoriasis Area and Severity Index score was 25. Complete blood count, liver/kidney function tests, and lipid profile were within normal ranges. Acitretin (25 mg/day) and topical calcipotriol-betamethasone ointment were prescribed.

The etiological relationship between psoriasis and vaccines is still uncertain. Immune dysregulation secondary to viral components and adjuvants of vaccines can cause the induction and exacerbation of psoriasis[2]. BCG has been used as a local immunotherapy for bladder cancer and can show its antitumor effects by starting an inflammation cascade resulting in the death of tumor cells. The side effects of BCG immunotherapy range from urinary tract symptoms to sepsis. Additionally, cutaneous findings following BCG immunotherapy, such as granulomatous skin lesions,

Address for Correspondence/Yazışma Adresi: Yusuf Can Edek MD, Gazi University Faculty of Medicine, Department of Dermatology, Ankara, Türkiye
E-mail: yusuf-can-35@hotmail.com **ORCID:** orcid.org/0000-0002-3877-8681
Received/Geliş Tarihi: 16.02.2024 **Accepted/Kabul Tarihi:** 09.09.2024 **Publication Date/Yayınlanma Tarihi:** 01.07.2025

Cite this article as/Atıf: Edek YC, Soygür A, Adışen E. Psoriasis exacerbation following intravesical BCG immunotherapy for bladder carcinoma: A case report. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):72-3

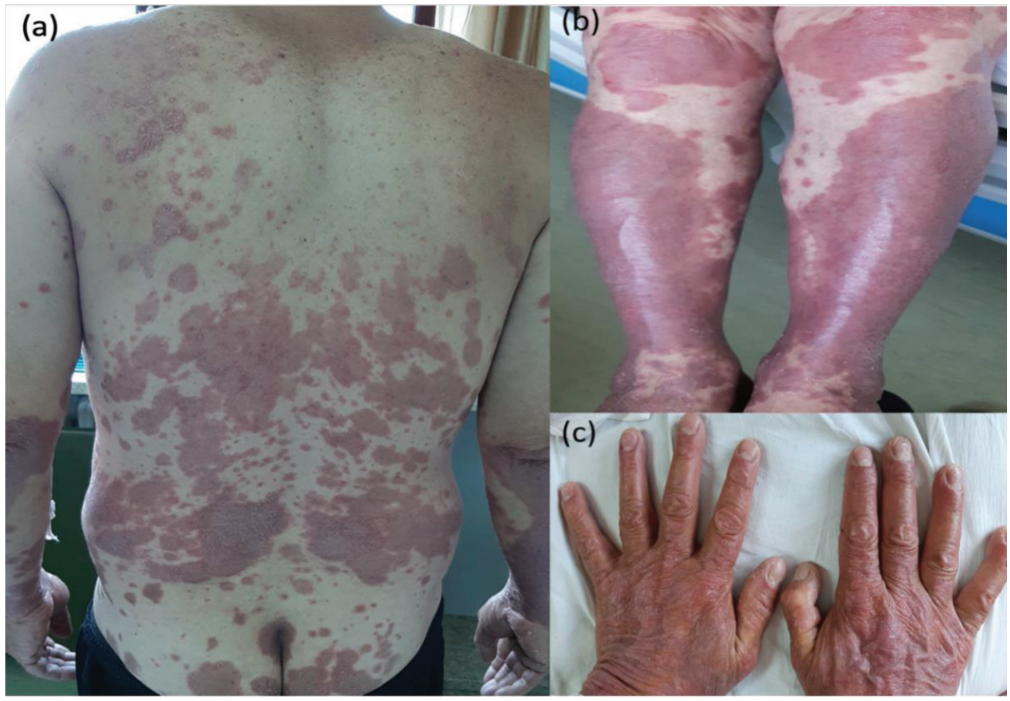

Figure 1. Widespread erythematous papules, plaques on the trunk **(a),** extremities **(b),** and pitting on the nails **(c)** one week after the third BCG immunotherapy

ulcers, and lymphadenopathy, have been reported[6]. BCG vaccine-induced psoriasis has been reported in a child[4]. Also, new-onset and flare-up of psoriasis-psoriatic arthritis cases following intravesical BCG immunotherapy were described in the literature[5-8]. Wee et al.[7] described a bladder carcinoma patient without psoriasis history under BCG immunotherapy who suffered erythrodermic pustular psoriasis. Queiro et al.[8] published a case that developed psoriatic arthritis following intravesical BCG application. BCG immunotherapy can induce Th1 and Th17-predominant immunologic response. Th1 and Th17 cells produce cytokines such as TNF-α, IL-12, IL17, IL-23, and IL-22 that have an important role in the psoriasis pathogenesis[2,9].

It is essential to remember that BCG immunotherapy can be a triggering factor for the exacerbation of psoriasis. With this case report, we would like to point out that the role of vaccines and immunotherapy in psoriasis pathogenesis should be taken into consideration and questioned. We think new case reports and studies will help us detect the relationship between psoriasis and BCG immunotherapy.

Ethics

Informed Consent: The patient in this manuscript has given written informed consent to the publication of his case details.

Footnotes

Authorship Contributions

Concept: Y.C.E., A.S., E.A., Design: Y.C.E., A.S., E.A., Data Collection or Processing: Y.C.E., A.S., E.A., Analysis or Interpretation: Y.C.E., A.S., E.A., Writing: Y.C.E., A.S., E.A.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Griffiths CE, Barker JN: Pathogenesis and clinical features of psoriasis. Lancet. 2007;370:263-71.
2. Gunes AT, Fetil E, Akarsu S, Ozbagcivan O, Babayeva L: Possible triggering effect of ınfluenza vaccination on psoriasis. J Immunol Res. 2015;2015:258430.
3. Macias VC, Cunha D: Psoriasis triggered by tetanus-diphtheria vaccination. Cutan Ocul Toxicol. 2013;32:164-5.
4. Koca R, Altinyazar HC, Numanoğlu G, Unalacak M: Guttate psoriasis-like lesions following BCG vaccination. J Trop Pediatr. 2004;50:178-9.
5. Dudelzak J, Curtis AR, Sheehan DJ, Lesher JL Jr: New-onset psoriasis and psoriatic arthritis in a patient treated with Bacillus Calmette-Guérin (BCG) immunotherapy. J Drugs Dermatol. 2008;7:684.
6. Larsen ES, Joensen UN, Poulsen AM, Goletti D, Johansen IS: Bacillus Calmette-Guérin immunotherapy for bladder cancer: a review of immunological aspects, clinical effects and BCG infections. APMIS. 2020;128:92-103.
7. Wee JS, Natkunarajah J, Moosa Y, Marsden RA: Erythrodermic pustular psoriasis triggered by intravesical bacillus Calmette-Guérin immunotherapy. Clin Exp Dermatol. 2012;37:455-7.
8. Queiro R, Ballina J, Weruaga A, et al.: Psoriatic arthropathy after BCG immunotherapy for bladder carcinoma. Br J Rheumatol. 1995;34:1097.
9. Lin Y, Slight SR, Khader SA. Th17 cytokines and vaccine-induced immunity. Semin Immunopathol. 2010;32:79-90.

Letter to the Editor
Editöre Mektup

DOI: 10.4274/turkderm.galenos.2024.23080
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):74-6

Self healing collodion baby: Case report

Kendi kendine iyileşen kollodiyon bebek: Olgu sunumu

Çağdaş Boyvadoğlu*, Fatih Tan, Hüseyin Serhat İnaloz****

*Ceyhan State Hospital, Clinic of Dermatology, Adana, Türkiye
**Gaziantep University Faculty of Medicine, Department of Dermatology, Gaziantep, Türkiye

Keywords: Collodion baby, self healing collodion baby, collodion phenotype

Anahtar Kelimeler: Kollodiyon bebek, kendi kendine iyileşen kollodiyon bebek, kollodiyon fenotip

To Editor,

Collodion baby (CB), describes a newborn phenotype in which the body surface is covered with a parchment-like membrane. Approximately 10% of colodion babies are observed to improve spontaneously within the first few weeks. This condition is referred to as "self-healing collodion baby" (SHCB) or "lamellar exfoliation of the newborn". Herein, we present a case of SHCB born with CB phenotype. A male newborn was born to a gravida 1 para 1 woman by cesarean section due to head-pelvis discordance. The baby was born as a term baby at 38 weeks of gestation, and there were no birth complications. The mother was 27 years old and had no known comorbidities except psoriasis. All prenatal laboratory values were within the normal range, and fetal ultrasound findings were normal. She received routine prenatal care and was followed up routinely during pregnancy. No consanguinity between mother and father was reported. There was no family history of genetic skin disease. The baby's birth weight was 2995 g, and his birth length was 45 cm. Appearance, pulse, grimace, activity, respiration scores were 7 and 9 at 1 and 5 minutes, respectively. Systematic examination of the baby at birth was normal. There were no signs of distal edema, sucking problems, or respiratory distress. No ear or hair abnormalities were detected, and no abnormal neurologic findings were found. On a dermatologic examination of the nose, fingers, and toes, especially covering the whole body, there was a parchment-like, shiny, taut membrane. There was diffuse erythema and scales all over the body. There were multiple fissures in the palms and flexural areas of the extremities (Figure 1a). Bilateral ectropion, eclabium, hypotrichosis of the eyebrows, and flattening of the nose were also present (Figure 1b). A limitation of movement in the joints due to the tense membrane was also observed. Serum electrolytes, protein, albumin, creatinine, fluid-electrolyte balance, and acute-phase reactants were closely monitored, and no abnormality was observed. Scrotal ultrasonography revealed increased fluid in the right scrotal cavity suggestive of a hydrocele. Abdominal ultrasonography and transfontanel ultrasonography were normal. The baby was kept in an incubator with humidity adjusted to 80% for 3 days, and vital values were closely monitored. His general condition was good during the follow-up. For skin care, the baby was bathed daily, and Hamamelis virginiana cream and emollient moisturizers (urea and salicylic acid-free) were used every 4 hours. Artificial tears were applied to keep the corneas moist. The baby was breastfed from birth.

Address for Correspondence/Yazışma Adresi: Hüseyin Serhat İnaloz MD, Gaziantep University Faculty of Medicine Department of Dermatology, Gaziantep, Türkiye
E-mail: serhatinaloz@hotmail.com **ORCID:** orcid.org/0000-0002-3860-9810
Received/Geliş Tarihi: 22.06.2024 **Accepted/Kabul Tarihi:** 09.09.2024 **Publication Date/Yayınlanma Tarihi:** 01.07.2025

Cite this article as/Atıf: Boyvadoğlu C, Fatih Tan F, İnaloz HS. Self healing collodion baby: case report. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):74-6

After 2 weeks of follow-up, the collodion membrane peeled off, leaving behind desquamation. A dermatologic examination two weeks later showed that the baby had a normal skin appearance except for desquamation in limited areas on the trunk (Figure 2). His eyes had a natural appearance. The patient was diagnosed as SHCB with the findings at follow-up, and the family was informed. At 6-month follow-up, the patient was observed to have a normal skin appearance (Figure 3).

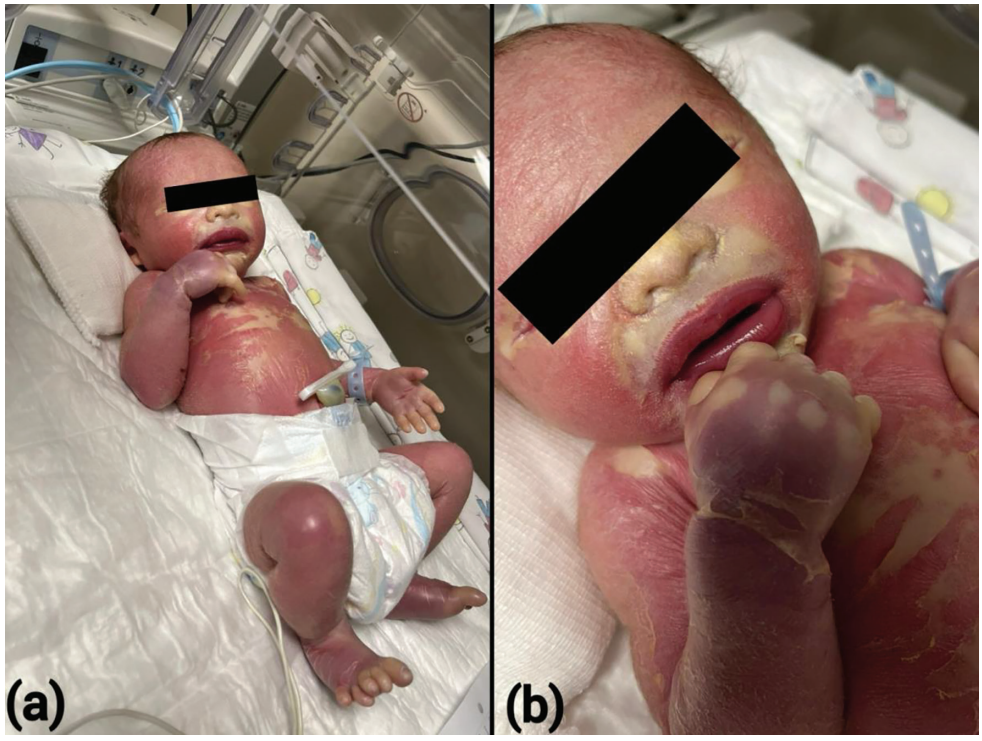

Figure 1. At birth **(a)** the whole body was covered with a parchment-like, shiny, and taut membrane, accompanied by diffuse erythema and scaling. **(b)** Bilateral ectropion, eclabium, hypotrichosis of the eyebrows, and nasal flattening were also observed

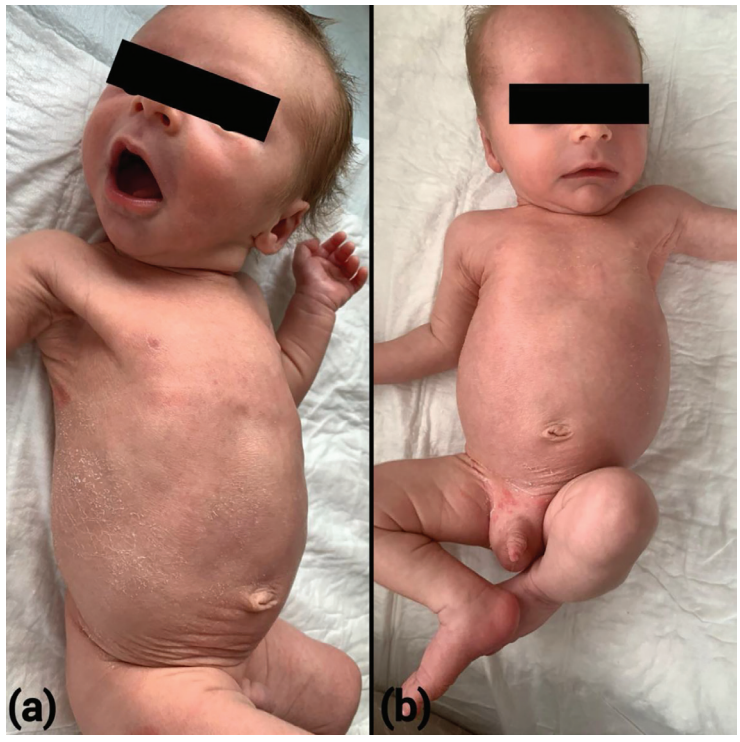

Figure 2. (a, b) Two weeks later, the baby showed normal skin appearance except for limited areas of desquamation on the trunk

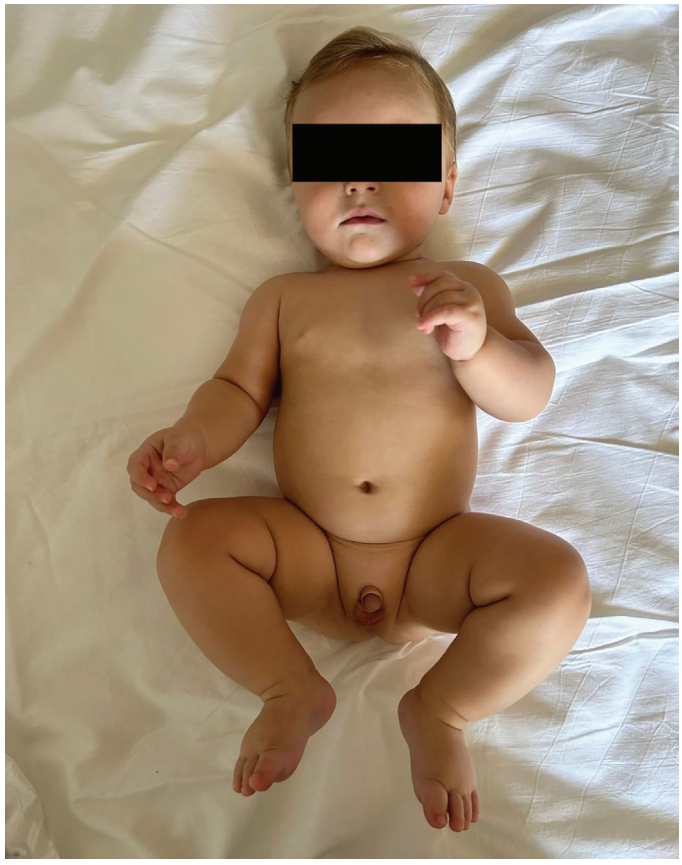

Figure 3. Presentation of completely normal skin at 6 months

The term CB was first described in 1884 by Hallopeau and Watelet. It can be seen in a range of diseases with varying clinical severity. The collodion phenotype is common to several different forms of autosomal recessive congenital ichthyoses[1].

Most CB are born at term and with a normal birth weight for the gestational week. It is estimated to occur in 1 in 50,000 to 100,000 births. CB is born with shiny, tight skin and erythroderma that resembles parchment covering the entire body, called the colodion membrane. After birth, ruptures and subsequent peeling of the inelastic membrane are observed. The membrane sloughs off in about 3-4 weeks and is usually followed by an ichthyosis phenotype. It is important to maintain fluid-electrolyte balance and infection control in infants born with the CB phenotype. Temperature control for the baby is important to reduce transepidermal water loss. Caution should be exercised with regard to the various complications that may occur, including hypernatremic dehydration, hypothermia, fissures, conjunctivitis, sepsis, constrictive bands, and edema of the extremities leading to vascular compromise. Patients with the CB phenotype should be provided with basic care support and close follow-up[2].

Some CB's improve spontaneously within the first few weeks and this has been termed SHCB[3]. Mutations in the *TGM1* gene encoding epidermal transglutaminase 1 have been associated with SHCB. Furthermore, mutations in the *ALOX12B* gene encoding 12(R)-lipoxygenase have also been shown to be related to SHCB[4,5]. In the follow-up of our patient, who was born with CB phenotype, the collodion membrane peeled off spontaneously, and completely normal skin appearance was observed except for desquamation. Our patient was diagnosed with SHCB based on clinical and follow-up findings. The limitation in our case management is that genetic testing could not be performed.

The birth of a baby with the CB phenotype is a worrying and difficult situation for the family. It is very important to provide psychological counseling to the family and to manage the process well.

Ethics

Informed Consent: Written informed consent to publish this case (including images) was obtained from the patient's mother.

Footnotes

Authorship Contributions

Surgical and Medical Practices: H.S.İ., Concept: Ç.B., H.S.İ., Design: Ç.B., H.S.İ., Data Collection or Processing: Ç.B., F.T., Analysis or Interpretation: Ç.B., F.T., H.S.İ., Literature Search: Ç.B., Writing: Ç.B.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Zdraveska N, Kostovski A, Sofijanova A, Jancevska S, Damevska K: Collodion phenotype remains a challenge for neonatologists: A rare case of self-healing collodion baby. Clin Case Rep. 2022;10:e6158.
2. Prado R, Ellis LZ, Gamble R, Funk T, Arbuckle HA, Bruckner AL: Collodion baby: an update with a focus on practical management. J Am Acad Dermatol. 2012;67:1362-74.
3. Chung M, Pittenger J, Tobin S, Chung A, Desai N: Expedient treatment of a collodion baby. Case Rep Dermatol Med. 2011;2011:803782.
4. Raghunath M, Hennies HC, Ahvazi B, et al: Self-healing collodion baby: a dynamic phenotype explained by a particular transglutaminase-1 mutation. J Invest Dermatol. 2003;120:224-8.
5. Vahlquist A, Bygum A, Gånemo A, et al: Genotypic and clinical spectrum of self-improving collodion ichthyosis: ALOX12B, ALOXE3, and TGM1 mutations in Scandinavian patients. J Invest Dermatol. 2010;130:438-43.

DOI: 10.4274/turkderm.galenos.2024.84829
Turkderm-Turk Arch Dermatol Venereol 2025;59(2):77-8

Lentigines confined to the upper body plaques of a paraplegic psoriasis vulgaris patient

Paraplejik bir psoriasis vulgaris hastasında vücudun üst yarısındaki plaklara sınırlı lentijinler

Gaye Güldiken Doğruel

İstanbul Arel University Faculty of Medicine, Memorial Atasehir Hospital, Clinic of Dermatovenereology, İstanbul, Türkiye

Keywords: Psoriasis, lentigines, lentiginosis, paraplegia, paralysis

Anahtar Kelimeler: Psoriasis, lentijinler, lentijinöz, parapleji, paraliz

To Editor,

Studies show that the active inflammatory environment in psoriasis vulgaris (PsoV) plaques reduces melanin production. In patients with moderate to severe PsoV, when one or more inflammatory molecules are inhibited during phototherapy or monoclonal antibody treatment, lentigines may develop on the healing plaques[1,2]. However, the development of lentigines in psoriasis patients who are not receiving treatment has not been reported. In this article, a paraplegic PsoV patient without any systemic psoriasis treatment, who developed lentigines on stable psoriasis plaques on the upper half of his body and his non-lesional oral mucosa, will be discussed.

A 65-year-old male presented to the dermatology clinic for psoriasis lesions. The patient, who has been paraplegic and wheelchair-bound for 40 years due to an electric shock, had a history of plaque-type psoriasis for 10 years, prostate cancer surgery 2 years ago, type-2 diabetes, and hypertension. He had been taking a candesartan/amlodipine combination, empagliflozin, acetylsalicylic acid for ten years, and leuprolide acetate as a gonadotropin-releasing hormone receptor (GnRHR) agonist since prostate cancer surgery. There was no history of medication use other than intermittent topical clobetasol propionate ointment for psoriasis. In the physical examination of the patient, there were melanocytic macular lesions on the edges and inner aspect of the lower lip, and melanocytic macules limited to the psoriatic plaques on both elbows, dorsum of the hands and umbilicus. These macules were absent on the PsoV plaques on both knees (Figure 1). The patient's Fitzpatrick skin type was 4. The patient claimed that lentigines appeared after the diagnosis of prostate cancer. There was no relevant pathology in the fingernails and toenails. The punch biopsy from the lentiginous lesion on the right elbow revealed psoriasiform hyperplasia and increased pigmentation in the epidermis.

Today, it has been revealed that there is an increase in tumor necrosis factor-alpha and interleukin-17 signals in PsoV, which synergistically cause an increase in the melanocyte number and a decrease in the pigmentation signal on psoriasis plaques[1]. Therefore, therapeutic inhibition of one of these pathways may lead to the elimination of the inhibitory effect on melanocytes in psoriasis plaques and an increase in pigmentation[1,2]. The fact that this patient was not receiving any systemic PsoV treatment while the

Address for Correspondence/Yazışma Adresi: Gaye Güldiken Doğruel MD, İstanbul Arel University Faculty of Medicine, Memorial Atasehir Hospital, Clinic of Dermatovenereology, İstanbul, Türkiye
E-mail: gayeguldiken@gmail.com **ORCID:** orcid.org/0000-0001-9582-7948
Received/Geliş Tarihi: 14.07.2024 **Accepted/Kabul Tarihi:** 22.10.2024 **Publication Date/Yayınlanma Tarihi:** 01.07.2025
Cite this article as/Atıf: Güldiken Doğruel G. Lentigines confined to the upper body plaques of a paraplegic psoriasis vulgaris patient. Turkderm-Turk Arch Dermatol Venereol. 2025;59(2):77-8

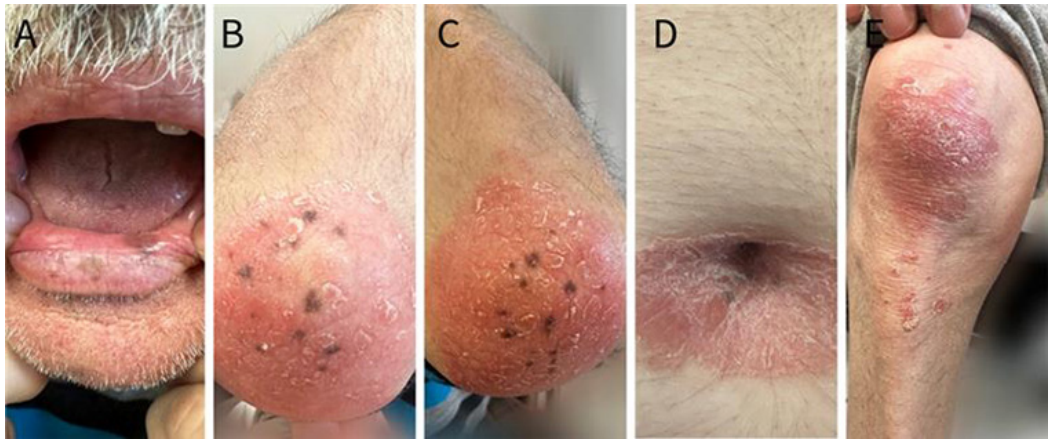

Figure 1. Clinical photographs of the patient. Lentigines on the lower lip **(A)**, lentigines confined to the psoriatic plaques on elbows and umbilicus **(B, C, D)**, the psoriasis plaque on the right knee without any lentiginous lesions **(E)**. (The backgrounds were blurred to hide the unnecessary details.)

lentigines appeared and that the lentigines formed on stable rather than healing plaques suggests that there may be additional triggering factors in the etiology.

Examining the patient's medication history, leuprolide acetate is a GnRHR agonist whose sex hormone-inhibiting effect is utilized in the treatment of prostate cancer[3]. In the literature, no increase in pigmentation has been reported in patients during GnRHR agonist treatments. Osuga et al.[4] reported no increase in skin pigmentation in 16 patients using GnRHR agonists. Therefore, it is unclear whether the leuprolide acetate was the trigger in our case. Besides, there are limited reports of amlodipine-associated pigmentation yet, and the reported hyperpigmentation was in a photo-distributed fashion, unlike the lentigines of our patient[5].

However, this sometimes, chronic application of topical immunomodulators might cause lentigines in chronic inflammatory plaque lesions, such as in the ILIAD phenomenon of atopic dermatitis[6]. Our patient's constant use of topical steroids might be the cause of lentiginosis. However, this does not explain the sparing of the lower body plaques perse.

Laugier-Hunziker syndrome is a sporadic lentiginosis of the oral mucosa and nails, which mostly appears in early and middle-aged adults, and is not associated with a systemic disease[7]. Such a condition might trigger the upper body PsoV plaques and lip pigmentation of our paraplegic patient; nevertheless, he had neither any nail pigmentation nor a history of neural, endocrine, or mesenchymal tumors, as in other precancerous lentiginosis syndromes[8].

Although there are not enough resources on how melanogenesis is affected in paraplegic patients, the fact that lentigines are not found on the lower extremity psoriatic plaques in this paraplegic patient suggest that neural factors may contribute. Further research is needed.

Ethics

Informed Consent: The patient's consent form including biopsy, photography, and scientific use was signed and kept in the Artvin Hopa State Hospital archives. The physical examination, biopsy, and construction of the article were all made by the author.

Footnotes

Financial Disclosure: The author declared that this study received no financial support.

References

1. Wang CQF, Akalu YT, Suarez-Farinas M, et al.: IL-17 and TNF synergistically modulate cytokine expression while suppressing melanogenesis: potential relevance to psoriasis. J Invest Dermatol. 2013;133:2741-52.
2. Di Cesare A, Fargnoli MC, Marinucci A, Peris K: Rationale for the development of speckled hyperpigmentation in the areas of psoriatic plaques after treatment with biologic agents. J Invest Dermatol. 2015;135:318-20.
3. Wilson AC, Meethal SV, Bowen RL, Atwood CS: Leuprolide acetate: a drug of diverse clinical applications. Expert Opin Investig Drugs. 2007;16:1851-63.
4. Osuga Y, Yano T, Kugu K, et al.: Effects of gonadotropin-releasing hormone analog treatment on skin condition. Gynecol Endocrinol. 2002;16:57-61.
5. Erbagci Z. Amlodipine associated hyperpigmentation. Saudi Med J. 2004;25:103-5.
6. Castelo-Soccio L, Di Marcantonio D, Shah P, Lee LW, Treat JR, Yan AC: Induced lentiginosis with use of topical calcineurin inhibitors. Arch Dermatol. 2012;148:766-8.
7. Lampe AK, Hampton PJ, Woodford-Richens K, Tomlinson I, Lawrence CM, Douglas FS. Laugier-Hunziker syndrome: an important differential diagnosis for Peutz-Jeghers syndrome. J Med Genet. 2003;40:e77.
8. Lodish MB, Stratakis CA: The differential diagnosis of familial lentiginosis syndromes. Fam Cancer. 2011;10:481-90.