Linear Atrophoderma of Moulin: A Case Report Highlighting Response to Methotrexate

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Abstract

Linear Atrophoderma of Moulin (LAM) is a rare dermatologic condition characterized by linear, hyperpigmented or hypopigmented atrophic lesions that follow Blaschko's lines. Typically emerging in childhood or early adulthood, Linear Atrophoderma of Moulin remains poorly understood, with few cases reported in the literature. It is often misdiagnosed due to clinical overlap with other dermatoses, particularly Linear Morphea. While the disease is generally benign, it can cause cosmetic and functional issues. No standardized treatment exists, and therapeutic responses vary.

We report the case of a 22-year-old female who presented with a 17-year history of unilateral hyperpigmented and mildly atrophic patches on her left side, including the arm, breast, trunk, and leg. These lesions followed Blaschko's lines and were accompanied by occasional calf pain and weakness. There was no history of inflammation, systemic symptoms, or relevant family history. Previous misdiagnoses included eczema and psoriasis, with no response to conventional treatments. A skin biopsy revealed basal layer hyperpigmentation, dermal collagen thickening, and perivascular lymphocytic infiltrates, supporting a diagnosis of Linear Atrophoderma of Moulin.

The patient was treated with oral methotrexate (20 mg weekly) and topical therapy consisting of betamethasone and calcipotriene. This treatment was maintained for one year under regular clinical monitoring.

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The patient showed significant clinical improvement, including reduced calf pain, diminished weakness, and noticeable fading of hyperpigmented lesions, no new lesions developed during treatment. However, three months after discontinuation of methotrexate, new lesions appeared and symptoms recurred, highlighting the potential need for ongoing management in chronic cases.

This case emphasizes recognizing Linear Atrophoderma of Moulin as a distinct clinical entity and differentiating it from mimicking conditions such as Linear Morphea. Methotrexate, in combination with topical corticosteroid and vitamin D analog therapy, appears to offer a promising treatment option. Continued research is essential to establish effective, long-term treatment protocols.

Keywords: Atrophoderma, Betamethasone, Blaschko's lines, Hypopigmentation, Hyperpigmentation, Methotrexate.

ضمور الجلد الخطي لمولان: تقرير حالة يسلط الضوء على الاستجابة للميثوتريكسات

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ملخص

ضمور الجلد الخطي لمولان (LAM) هو حالة جلدية نادرة تتميز بآفات ضمورية خطية، أو مفرطة التصبغ، أو ناقصة التصبغ، تتبع خطوط بلاشكو. يظهر ضمور الجلد الخطي لمولان عادةً في مرحلة الطفولة أو البلوغ المبكر، ولا يزال غير مفهوم جيدًا، مع وجود حالات قليلة مسجلة في المراجع الطبية. غالبًا ما يُشخص خطأً بسبب التداخل السريري مع أمراض جلدية أخرى، وخاصةً القشيعة الخطية. على الرغم من أن المرض حميد بشكل عام، إلا أنه قد يسبب مشاكل تجميلية ووظيفية. لا يوجد علاج موحد، وتختلف الاستجابات العلاجية.

نُبلغ عن حالة امرأة تبلغ من العمر 22 عامًا، حضرت إلى المستشفى بتاريخ 17 عامًا من بقع أحادية الجانب مفرطة التصبغ وضامرة قليلاً على جانبها الأيسر، بما في ذلك الذراع والثدي والجذع والساق. اتبعت هذه الآفات خطوط بلاشكو، وكانت مصحوبة بألم وضعف في الساق من حين لآخر. لم يُسجل أي تاريخ التهابي، أو أعراض جهازية، أو تاريخ عائلي ذي صلة. وشملت التشخيصات الخاطئة السابقة الإكزيما والصدفية، دون استجابة للعلاجات التقليدية. كشفت خزعة الجلد عن فرط تصبغ في الطبقة القاعدية، وسماكة في الكولاجين الجلدي، وتسللات لمفاوية حول الأوعية الدموية، مما يدعم تشخيص ضمور الجلد الخطي لمولين.

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عولج المريض بمزيج من الميثوتريكسات الفموي (20 ملغ أسبوعيًا) والعلاج الموضعي المكون من بيتاميثازون وكالسيبوتريين. استمر هذا العلاج لمدة عام تحت المراقبة السريرية المنتظمة.

أظهر المريض تحسنًا سريريًا ملحوظًا، بما في ذلك انخفاض ألم الساق، وانخفاض الضعف، وتلاشي ملحوظ للآفات المفرطة التصبغ. لم تظهر أي آفات جديدة أثناء العلاج. ومع ذلك، بعد ثلاثة أشهر من إيقاف الميثوتريكسات، ظهرت آفات جديدة وعادت الأعراض، مما يُبرز الحاجة المحتملة إلى العلاج المستمر في الحالات المزمنة.

تُؤكد هذه الحالة على أهمية التعرف على ضمور الجلد الخطي لمولان كحالة سريرية مميزة، وتمييزه عن الحالات المشابهة له مثل القشرية الخطية. يبدو أن الميثوتريكسات، مع الكورتيكوستيرويدات.

الكلمات المفتاحية: ضمور الجلد، بيتاميثازون، خطوط بلاشكو، نقص التصبغ، فرط التصبغ، ميثوتريكسات.

Introduction:

Linear Atrophoderma of Moulin (LAM) is a rare, progressive dermatological disorder characterized by linear atrophic lesions that can be localized or generalized. These lesions often present as hyperpigmented or hypopigmented depressed scars (Moulin G et al.,1992). The exact etiology remains uncertain, though inflammatory and immunological mechanisms are implicated (Danarti R et al., 2003). Only a few hundred cases have been documented (Tan SK et al., 2016), with most individuals presenting in childhood or early adulthood (Norisugi O et al.,2011). The condition demonstrates a slight female predominance (Cecchi R et al.,1997), and while some cases are familial (López et al., 2008), most are sporadic (Baumann et al. , 1994). Lesions typically start as small areas of atrophy, often on the limbs or face, and may progress slowly, leading to cosmetic and functional impairment (Wollenberg A et al. , 1996) Although LAM itself is not life-threatening, it has significant psychological and physical consequences, especially when associated with musculoskeletal complications, such as joint contractures or muscle wasting (Artola Igarza JL et al. ,1996). Prognosis varies, with many patients experiencing stable, localized disease, while generalized forms may result in long-term disability (Browne C et al.,2000). Treatment is symptomatic, focusing on managing the skin lesions with topical therapies and physical rehabilitation for musculoskeletal issues (Rompel et al., 2000). Systemic involvement is rare, and mortality is uncommon (Martin L et al., 2002).

LAM, first described by Moulin et al. (1992), is an enigmatic skin disorder characterized by increased pigmentation in the form of slightly atrophic patches that follow the lines of Blaschko (Utikal et al., 2003). Despite being described in the literature for over two decades, only a few isolated cases have been reported (Tan SK et al., 2016; Zampetti A et. al., 2008). However, some with the initial description (Larregue et. al.,1995). In this study, we present a case that exhibited

typical clinical and histopathologic features of LAM and conducted a therapeutic trial to assess treatment efficacy and its limitations to improve patient outcomes and guide future research. Although the exact pathophysiology and etiology remain poorly understood, current evidence suggests a role for postzygotic mosaicism and immune-mediated mechanisms (Danarti et. al., 2003; Ang et. al., 2005), particularly due to its distribution along Blaschko's lines and histopathological findings such as basal layer hyperpigmentation and superficial perivascular lymphocytic infiltrates (Ang et. al., 2005). A brief review of the literature highlights the absence of

standardized treatment protocols (Zhang et. al,2020), though several therapeutic options have been explored with variable success (Villani et. al., 2013).

Patient Background:

A 22-year-old female presented with a 17-year history of unilateral, hyperpigmented brown patches initially appearing on her left arm, upper breast, trunk, and left leg, with mild pain in the calf area. The lesions progressively spread despite no signs of inflammation, erythema, or edema (Miteva et. al., 2005). The patient also reported occasional weakness in her left lower limb, particularly in the calf region, but no significant family or medical history was noted (López et. al.,2008). Her condition had been misdiagnosed as eczema and psoriasis in early years (Ripert et. al.,2010), with treatments such as topical creams and laser therapy failing to improve the lesions (Schepis et. al., 2010).

Presentation:

On examination, the patient exhibited linear hyperpigmentation, mild atrophic changes, hypopigmentation, and shiny skin with slight depression on palpation, mainly on the left calf, upper thigh, and loin. New lesions showing prominent atrophy were detected on the left breast and arm, which followed Blaschko's lines along the same leg. Neurological examination revealed mild, intermittent calf pain and hyperreflexia in the contralateral leg. However, a full neurological workup was not pursued due to lack of additional signs. The patient was counseled on monitoring for neurological symptoms and remains eligible for neurology referral if her condition changes.

Diagnostic workup:

Laboratory investigations, including complete blood count, liver and renal profiles, and antinuclear antibodies, returned normal results (Nanchaipruek et. al.,2024).

Histopathologic findings: An incisional biopsy from one of the hyperpigmented, atrophic patches on the left calf showed a mildly thinned epidermis with flattening of the ridges, prominent basal layer hyperpigmentation, and mild superficial perivascular lymphocytic infiltrate (Norisugi et. al.,2011; Di Lernia et. al., 2011). In the dermis, subtle thickening and homogenization of collagen bundles were noted without dermal sclerosis, distinguishing LAM from linear morphea. (Ang et. al., 2005; Bolognia et. al.,

2012) The subcutaneous fat layer was unremarkable, further differentiating LAM from sclerosing dermatoses (Norisugi et. al., 2011).

Other investigations: Immunohistochemistry showed sparse T-lymphocyte predominance and preserved basement membrane integrity, with no immune complex deposition on direct immunofluorescence (Zouboulis et. al, 2012).

Given the clinical and histological findings, a diagnosis of LAM was made, and the patient was treated with methotrexate (20 mg weekly) and a topical combination of betamethasone and calcipotriene. Over one year, calf pain resolved, weakness improved, hyperpigmentation reduced, and no new lesions appeared. However, three months after methotrexate cessation, symptoms recurred, suggesting a potential need for maintenance therapy (Vasquez MA et al., 2015) .

Discussion:

The diagnostic criteria for LAM include childhood or adolescent onset, unilateral hyperpigmented, slightly atrophic lesions along Blaschko's lines on the limbs or trunk, absence of inflammation or induration, and a stable course (Kaur et. al., 2016). Histopathologically, LAM features hyperpigmentation of the basal epidermis and otherwise normal connective tissue (Ang et. al.,2005). It has been reported across various demographics (López et. al., 2008).

The classification of LAM remains debated. Some consider it within the morphea spectrum (Bolognia et. al., 2012), while others argue for its recognition as a distinct entity due to absent sclerosis, systemic features, and its unique presentation (Ang et. al., 2005). Ongoing molecular and immunological research is crucial for defining its pathogenesis and management (Norisugi et. al., 2011; Chavez et. al., 2021).

LAM should be differentiated from linear morphea as the two differ in prognosis and clinical features. LAM typically has a better prognosis due to the absence of sclerosis. While no definitive cure exists, several treatments have been explored:

1. Topical Calcipotriol: Shown to improve lesions in a 19-year-old female (Kaur et. al., 2016). Potassium Aminobenzoate (Potaba): Early study have suggested clinical benefits (Artola Igarza et. al.,1996).
2. Methotrexate: Demonstrated success in several cases (Norisugi et. al.,2011; Vasquez et. al., 2015).
3. Intralesional Platelet-Rich Plasma (PRP): Modest improvement in plaque depth reported (Kaur et. al., 2016).

The classification of LAM as a distinct condition versus part of a spectrum including Atrophoderma of Pasini and Pierini or linear scleroderma remains under investigation (Ang et. al.,2005; Zouboulis et. al., 2012).

In conclusion, Linear Atrophoderma of Moulin remains a poorly understood condition, often misdiagnosed due to its clinical similarities with Linear Morphea (Table1). However, its characteristic distribution along Blaschko's lines, unilateral pattern, favorable prognosis, and promising response to methotrexate therapy emphasize the need for further research to understand its pathophysiology better and optimize treatment strategies.

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Ethical approval: IRB approval number: (1562024), Date: (15-5-2024)
Consent (written consent is submitted with the research)

Authors' contributions:

Conceptualization:	Researcher1
Writing -original draft:	Researcher1
Writing -review and editing:	Researcher2
History + physical examination (including biopsy +dermoscopy):	Researcher1+ Researcher2
Investigation:	Researcher1+ Researcher2
Data collection:	Researcher1
Management and documentation of results:	Researcher1+ Researcher2
IRB preparation and communication to the journal:	Researcher3
Project administration:	Researcher3
Supervision:	Researcher3

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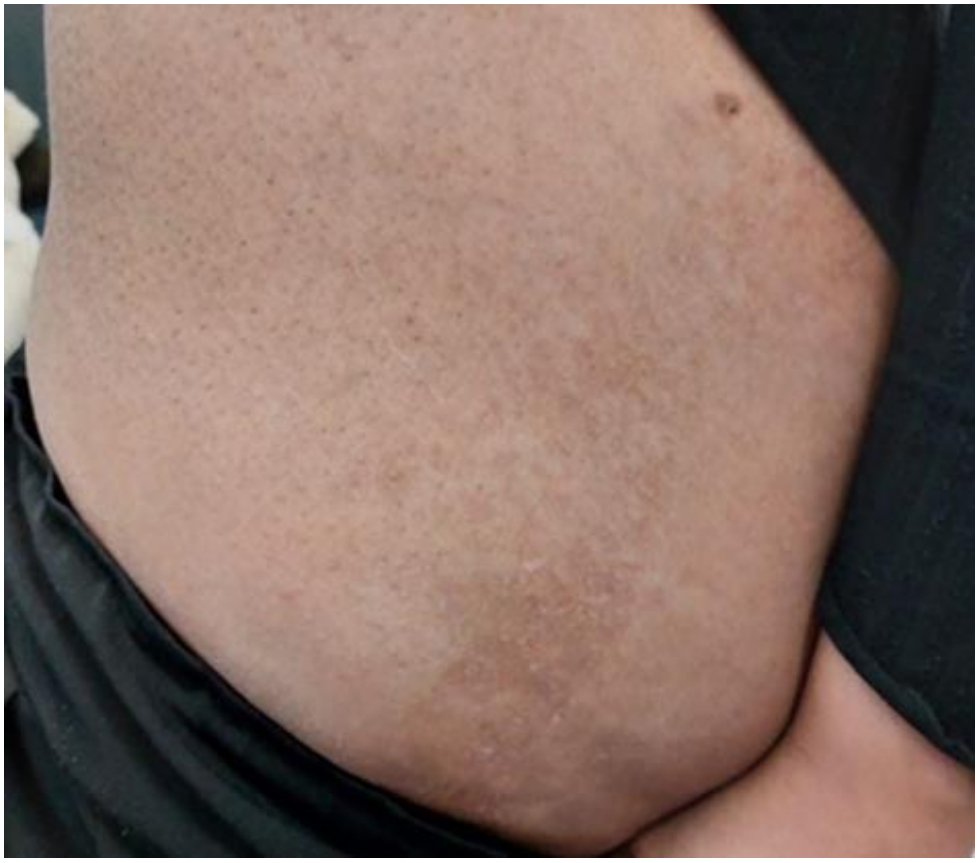
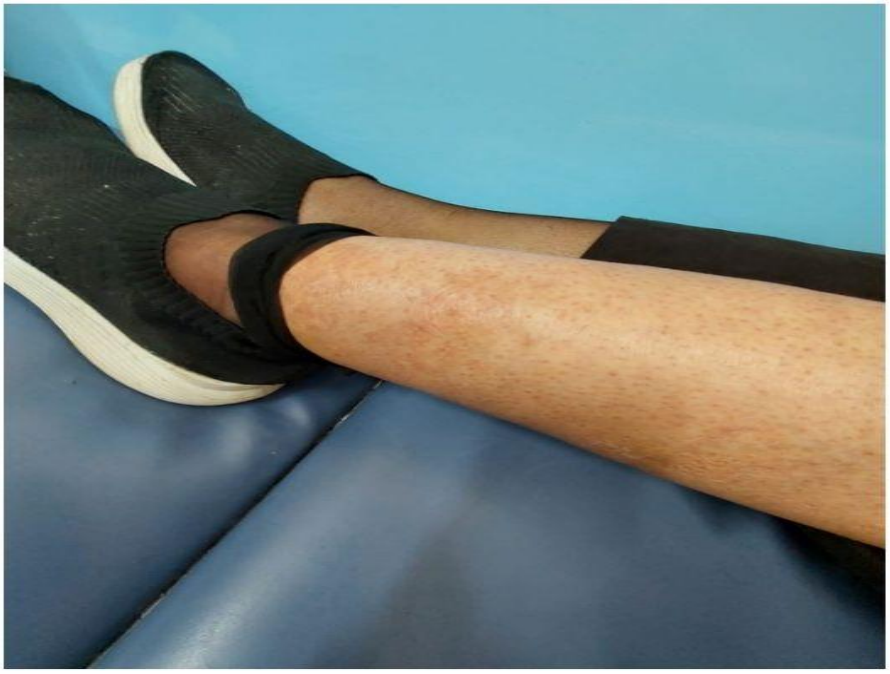
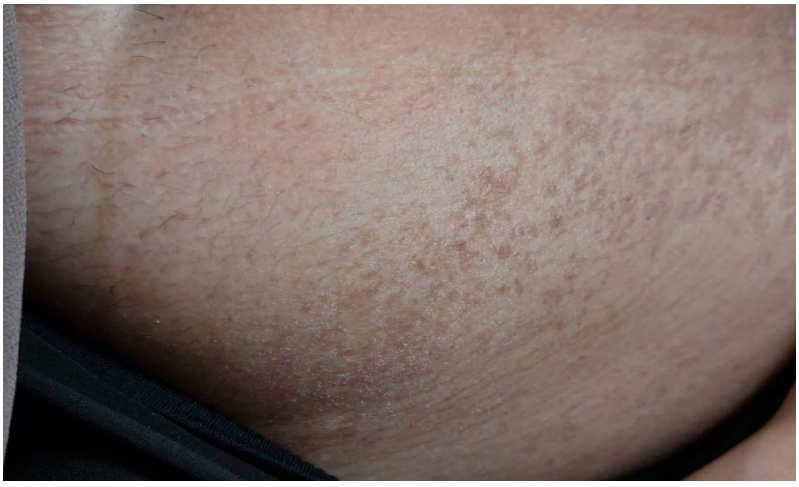


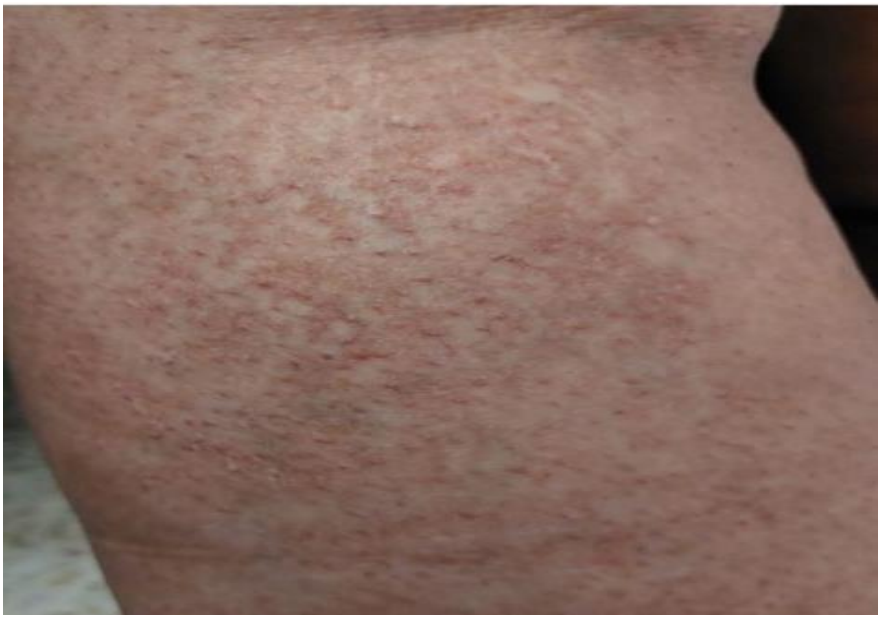
(Figure1: This figure shows the linear hyperpigmentation, mild atrophic hyperpigmentation, hypopigmentation, and shiny skin patches that were present on our patient's left upper thigh and trunk.)



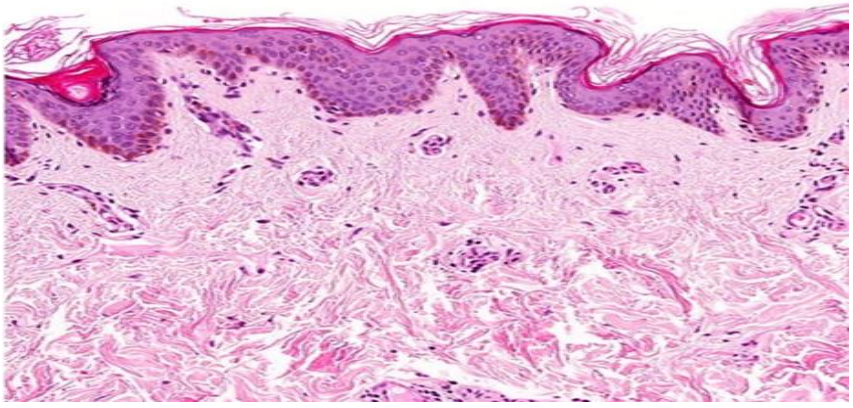
(Figure2:This figure shows the linear hyperpigmentation, mild atrophic hyperpigmentation, hypopigmentation, and shiny skin) patches that were present on our patient's left calf)



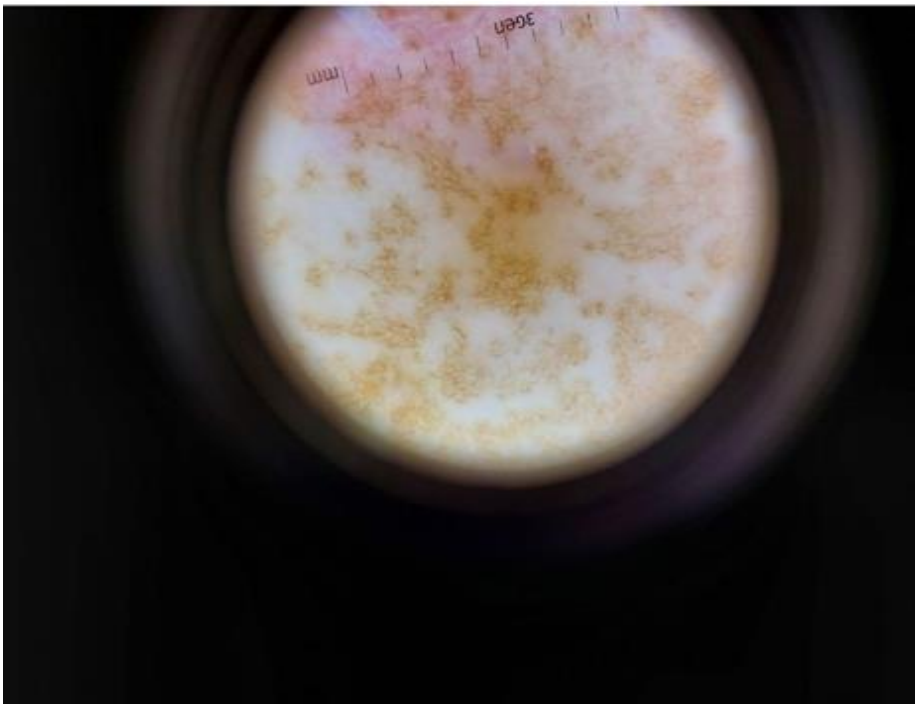
(Figure3: This figure shows hyperpigmented patches on the left loin)



(Figure4: This figure shows hyperpigmented brown patches on the left calf with shiny, dry skin features.)



(Figure5: This figure shows a typical microscopic configuration of a biopsy for linear Atrophoderma of Moulin, which was similar to our results.)



(Figure6: This figure shows how the skin looks under a dermoscopy)

Table (1): Comparison between Linear Atrophoderma of Moulin and Linear Morphea

Features	Linear Atrophoderma of Moulin (LAM)	Linear Morphea
Age of onset	Usually in childhood or early adulthood	Common in children, but can occur at any age
Distribution	Follows blaschko's lines(typically unilateral)	May follow blaschko's lines, also commonly unilateral
Appearance of lesions	Hyperpigmented or hypopigmented .slightly atrophic patches with no induration	Indurated, sclerotic plaques that may become hypopigmented or hyperpigmented

Progression	Slowly progressive or stable, non-destructive	Often progressive. May lead to tissue fibrosis and deformity
Symptoms	Usually asymptomatic, occasional mild pain or weakness	May be associated with tightness, discomfort or joint limitations
Inflammation	Absent or minimal	Often present, especially in early inflammatory phase
Epidermal changes	Mild perivascular lymphocytic infiltrate ,no significant immune deposits	May show epidermal atrophy or hyperkeratosis
Dermal findings	Thickened collagen bundles ,no sclerosis or fibrosis	Prominent dermal sclerosis ,loss of adnexal structures
Subcutaneous fat	Typically preserved	Often shows fat loss and deeper tissue involvement
Immunohistochemistry	Mild perivascular lymphocytic infiltrate , so significant immune deposits	Inflammatory infiltrates, may show autoantibody presence
Systemic involvement	Rare	Can be associated with extracutaneous manifestations
Prognosis	Benign , cosmetics concerns dominate	Variable can cause functional impairment if untreated
Response to treatment	Variable may show improvement with methotrexate or topical agents	Often require systemic immunosuppression(Ex. Methotrexate, Corticosteroids)

Ethical Approval:

Mutah University

Faculty of Medicine

Ethics Committee

Professor Eman Albataineh
Chairman of the Ethics Committee
Faculty of Medicine
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Reference Number: 1562024

Review Report

Dear Dr. Amal Aqeel Odeh Al -btoosh

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The Ethics Committee in the faculty of medicine reviewed your project with the title " **A case of linear atrophoderma of Moulin."**

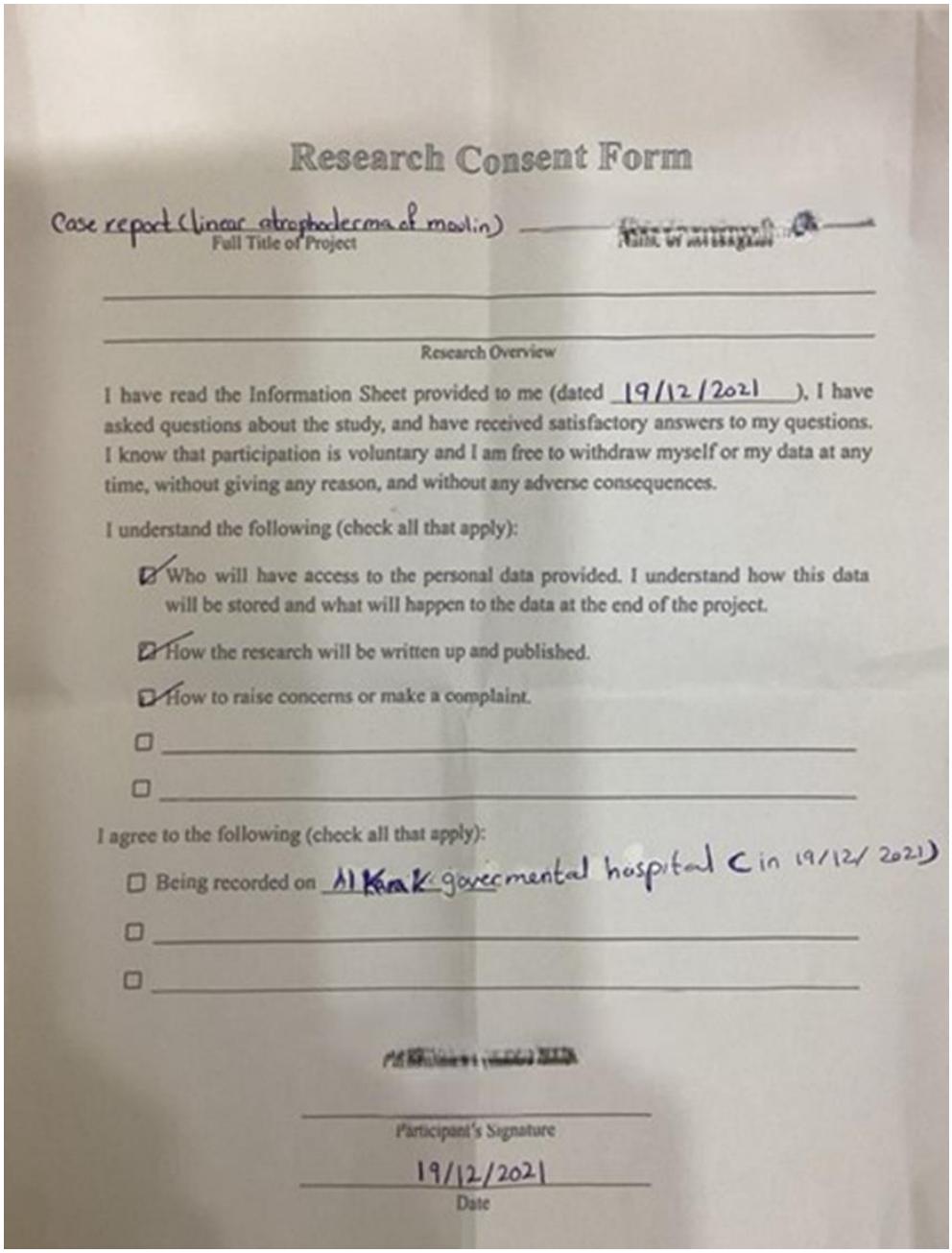
Based on our ethical considerations and guidelines, the committee has approved your project without any amendments.

Chairman of the Ethics Committee

15-5-24

Mu'tah University Faculty Of Medicine

Consent form:



Research Consent Form

Case report (linear atrophoderma of maolin)
Full Title of Project

[redacted]

Research Overview

I have read the Information Sheet provided to me (dated 19/12/2021). I have asked questions about the study, and have received satisfactory answers to my questions. I know that participation is voluntary and I am free to withdraw myself or my data at any time, without giving any reason, and without any adverse consequences.

I understand the following (check all that apply):

- ☑ Who will have access to the personal data provided. I understand how this data will be stored and what will happen to the data at the end of the project.
- ☑ How the research will be written up and published.
- ☑ How to raise concerns or make a complaint.
- ☐ _____
- ☐ _____

I agree to the following (check all that apply):

- ☐ Being recorded on Al Karak gavecmental hospital C in 19/12/2021)
- ☐ _____
- ☐ _____

[redacted]

Participant's Signature

19/12/2021
Date

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