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Original Article

Photosensitivity and calcinosis cutis are the major clinical presentations in patients with juvenile and adult dermatomyositis

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Abstract

Background Dermatomyositis is an uncommon inflammatory disease that causes muscle weakness and a distinctive dermatological rash. The objective of this study is to record all clinical presentations in patients with dermatomyositis but mainly dermatological manifestations.

Methods This case series descriptive study included 23 patients with dermatomyositis who were seen during the period from 2014-2023. A full history and clinical examination were carried out. All patients were referred from the Rheumatology and Pediatric Units after doing a full assessment and necessary investigations.

Results The juvenile group peak consisted of 13 patients, their ages ranged from 3-15 years with a mean of 5 years, 6 males and 7 females. The main clinical picture was photosensitivity with distinctive heliotrope facial rash in all patients, and calcinosis cutis in 8 (61.53%) cases The adult group included 10 subjects, their ages ranged from 30-60 years with a mean of 47 years, 9 females and one male patient. The presenting picture was skin rash in a form of photosensitivity with characteristic heliotrope of the face and muscle weakness of shoulder and pelvic girdles in all cases. No malignancy was reported.

Conclusion There was no significant difference between both genders in children, however, it was more prevalent in adult females. The presenting features in the juvenile group were photosensitivity, a heliotrope rash of the face and calcinosis cutis. While photosensitivity with heliotrope dermatitis of the face and muscle weakness was sever in adults. No malignancy was detected during follow-ups in any patient.

Key words
Photosensitivity; Calcinosis cutis; Dermatomyositis; Heliotrope Facial rash; Muscle weakness.

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Introduction

Dermatomyositis (DM) is a systemic, autoimmune, idiopathic, progressive inflammatory disease of the small vessels, mostly affecting the muscles and skin, characterized by myopathy and specific cutaneous manifestations. DM has several phenotypes, like myositis, skin reaction, and interstitial lung disease.[1]

All ethnic groups are affected and can occur at

any age. DM is classified according to age, Juvenile DM (JDM): affects children younger than 18 years old, affecting nearly three in one million per year, and girls are affected around twice as frequently as boys. Adult DM (ADM): disease experienced in those older than 18 years, at age around 40-50 years. The incidence is approximately 9.63/ million adults per year.[2-4]

Cutaneous findings of DM include Gottron sign which appear as pink papules on the metacarpophalangeal and interphalangeal joints, scaly erythema involving the photo-exposed parts, heliotrope rash as periorbital violaceous erythema and edema, and periungual telangiectasis with hypertrophy of cuticle.[5-7] Additionally vasculitis, panniculitis, localized lipoatrophy, skin ulcers, and poikiloderma.[8,9] Recently, pityriasis rubra pilaris-like eruption were reported as an uncommon skin manifestation in DM.[10]

Calcinosis cutis is characterized by soft tissue calcification with abnormal calcium deposition in the skin, subcutaneous tissue, and muscle. Calcinosis cutis is considered a rare condition and it has 5 different types. Dystrophic calcinosis is the primary one and it is linked with a variety of autoimmune disorders like DM.[11,12] Dystrophic calcinosis is sub-classified into 4 overlapping forms: calcinosis circumscripta (apparent papules or nodules around joints and on the skin), tumoral calcinosis (arises deeper in the dermis, subcutaneous tissue, fascia, and/or muscle), calcinosis universalis (myofascial planes deposition), and exoskeletal calcinosis (generalized deposition).[13]

Nearly 30 years ago, Bohan *et al.*[5] published the diagnostic criteria for DM. They used five analytical criteria: distinctive skin rash, weakness of the proximal muscles, elevated muscle enzymes, the electromyogram (EMG) finding of myopathic changes, and characteristic muscle biopsy. Recognition of cutaneous manifestations in DM is crucial for rapid and accurate diagnosis and further treatment of DM.[13]

This present study is mainly concerned with cutaneous manifestations as their presence has important diagnostic value in dermatomyositis. They are easily detected and evaluated without a need for further investigations, especially in chronic cases where muscle enzymes went to normal level.

Patients and methods

This is a case series descriptive study where 23 patients with dermatomyositis were seen at a private clinic (KES) and at Baghdad Teaching Hospital, Baghdad, Iraq during the period from 2014-2023. A full history and demographic features were recorded and a thorough clinical examination and evaluation were carried out. All patients were referred from the Rheumatology and Pediatric Units after doing full assessment and necessary investigations. The diagnosis was made rendering to the indicative criteria defined by Bohan *et al;* 1975.[5]

Juvenile DM included children of both genders younger than 18 years, while adult DM involved those older than 18 years. Plan X- radiography to document nodular calcinosis lesions was arranged. Almost all of the participants' skin was classified as Fitzpatrick types III or IV. Approval was obtained from the Ethical Committee after a full explanation of the nature of the study and publication of data and digital photographs. Written informed consent was obtained from the participants or their parents.

Results

Twenty-three patients with an age range of 3-60 years were assessed, 15 (65.2%) females and 8 (34.8%) males, with a male-to-female ratio of 1:3. These patients were classified into juvenile and adult groups.

Table 1 The frequency rate of skin manifestations in JDM and ADM.

Skin manifestations	*(JDM; n=13)* *N (%)*	*(ADM; n=10)* *N (%)*
Photosensitivity	13 (100%)	10 (100%)
Heliotrope rash with periorbital edema	12 (92%)	10 (100%)
Facial violate scaly papules and macules- face mask	5 (38.5%)	9 (90%)
Facial erythema- butterfly distribution	5 (38.5%)	3 (30%)
Symmetrical red scaly papules and macules of the forearms	1 (7.7%)	2 (20%)
Violaceous scaly papules and macules of the V-neck	0	3 (30%)
Gottron papules/sign	2 (15.4%)	2 (20%)
Periungual erythema and telangiectasias	0	1 (10%)
Calcinosis cutis	13 (100%)	1 (10%)
Upper limbs	12 (92%)	1 (10%)
Lower limbs	9 (69.2%)	0
Buttock	4 (30.8%)	0
Pyoderma and ulceration over calcification	6 (46.2%)	0
Bilateral Mechanic's hands	3 (23.1%)	1 (10%)
Multiple facial ulcers	5 (38.5%)	0

JDM: juvenal dermatomyositis, ADM: adult dermatomyositis.

The JDM consisted of 13 patients, whose ages ranged from 3-15 years with a mean of 5 years, 6 (46.2%) boys and 7 (53.8%) girls. The main clinical cutaneous picture was photosensitivity with distinctive heliotrope facial rash in all patients. They exhibited erythematous scaly patches over the sun-exposed portions of the face, and sometimes the upper limbs, sparing the chin, back of the ears, the legs, and feet **(Figure 1)**. Physical examination revealed periorbital edema with pink purple color over the eyelids (heliotrope color rash) in 12 (92%) juvenile cases.

Five (38.5%) patients showed mask-like facial violate scaly papules and macules. Similar erythema sometimes occurred only over the malar prominences in 5 (38.5%) participants **(Figure 2A)**. Few patients experienced the Gottron papules/sign which is characterized by violaceous papules and erythema involving the extensor surfaces of the interphalangeal joints and knuckles, or over the elbows or knees. Multiple small facial ulcers were presented in 5 (38.5%) cases (**Table 1**). Calcinosis cutis was another major complaint, which was experienced in 8 (61.5%) JDM individuals and was characterized by multiple hard tender nodules and sheet like subcutaneous plaques present on the upper limbs, lower extremities,

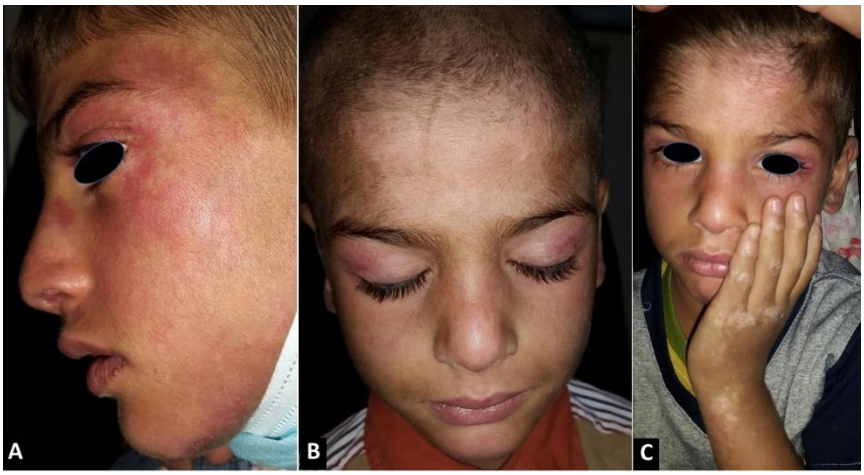

Figure 1 Showing three children with photosensitivity rash with periorbital heliotrope rash and edema, (A, B & C), Gottron papules and macules (C).

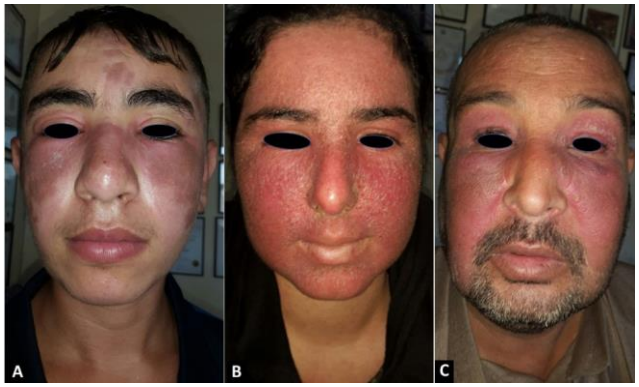

Figure 2 A boy with DM complain of periorbital edema and heliotrope rash associated with butterfly-like facial violate scaly papules (A), Two ADM had face mask violate scaly papules and macules and heliotrope rash (B&C).

and buttock in 12 (92%), 9 (69.2%), 4 (30.8%) patients, respectively **(Figure 3)**. Six (46.2%) of the participating children revealed overlying pyoderma and ulceration **(Figure 4)**. They occurred in several locations such as the thigh, knees, elbows, hands and digital pulps. Whenever there was pyoderma, ulceration and scar, there was underlying calcinosis and calcification. Mechanic's hands, defined as asymptomatic calcified hyperkeratotic papules, cracking (psoriasis-like lesions) of both hands, occurred in 3 (23.1%) subjects **(Figures 3C&4C)**. X-ray of the involved portion of calcinosis cutis showed apparent calcification within subcutaneous soft tissues. All patients complained of muscle weakness and atrophy of the pelvis girdle.

Whereas the ADM group peak included 10 patients, their ages ranged from 30-60 years with a mean of 47 years. There were 9 (90%) females and one (10%) male patient. The presenting picture was a skin rash in the form of severe recalcitrant photosensitivity over the sun-exposed parts. All subjects with ADM revealed heliotrope rash with periorbital edema, while 9 (90%) patients had the characteristic facial violate scaly papules and macules (heliotrope face mask) **(Figure 2B&C)**. There were erythematous scaly papules and macules involving the V-neck and the forearms in 3 (30%) and 2 (20%) cases, respectively **(Figure 5)**. The Gottron papules/ sign was exhibited in 2 (20%) individuals. Periungual erythema and telangiectasias were reported in 1 (7.7%) case.

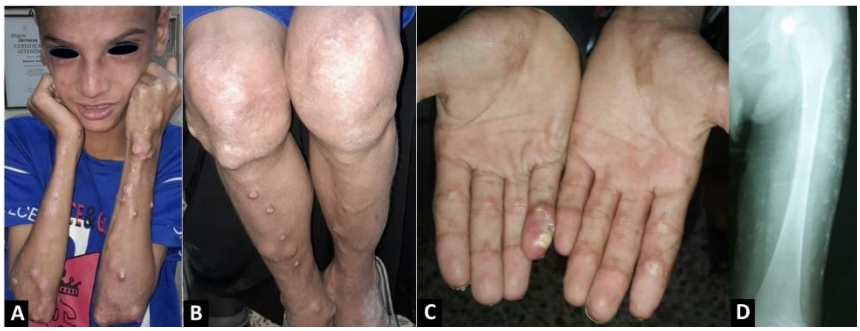

Figure 3 Showing a boy with JDM experienced calcinosis cutis which characterized by multiple hard tender nodules, involving forearms and legs (A&B), Bilateral Mechanic's hands (C), plan X-rays showing multiple deep calcifications of the left thigh.

Figure 4 Showing a girl with JDM revealed multiple facial ulcers as a part of heliotrope (A), calcinosis cutis associated with pyoderma and ulceration involving forearms, buttock, and legs (B, D, &E), Mechanic's hand (C).

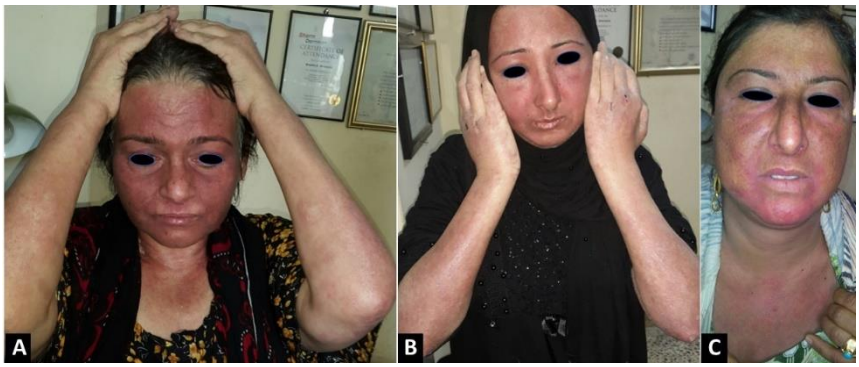

Figure 5 Showing 3 women with ADM with photosensitivity revealed periorbital edema heliotrope rash. The condition associated with symmetrical red scaly papules and macules of the forearms (A&B), violaceous scaly papules and macules of the V-neck (A&C).

Calcinosis cutis was revealed in one (10%) crippled female patient, it was seen around the elbows and accompanied by Mechanic's hands. The adult variant of DM was associated with muscle weakness of the shoulder and pelvic girdles in all patients. No malignancy was seen throughout the disease course.

Discussion

The present work is mainly concerned with cutaneous manifestations as they are the more obvious early clinical signs that facilitate the right clinical diagnosis, both in JDM and ADM.

Dermatomyositis chiefly affects the muscles and skin through the microangiopathic condition, the disease has a bimodal pattern, juvenile and adult variants.[1] Patients of DM have a diversity of cutaneous findings which are significant components in the evaluation of both current disease activity and long-lasting lesions associated with damage.[14]

Each patient was examined and studied in the Rheumatology or Pediatric Units and full assessment and necessary investigations suitable for the DM were done. Thereafter, patients were referred to the Dermatology Unit.

The frequency of increased cutaneous photosensitivity in DM patients has been poorly recognized. Many previous studies found different cutaneous findings of DM including Gottron papules/ sign, scaly erythema involving the photo-exposed parts, heliotrope rash, and periungual telangiectasis with hypertrophy of the cuticle.[5-7] Other preceding documents reported rare skin diseases such as calcinosis cutis, vasculitis, panniculitis, localized lipoatrophy, poikiloderma, alopecia, and PRP-like eruption.[8-10] Thus, documentation of all specific dermatological manifestations associated with DM could be considered as a main noninvasive visible criterion of DM and can help in early discovery, assessment and rapid management of the disease.

The present work indicates that females were more commonly affected by DM than males, with a male to female ratio of 1:3. While showing no gender difference during the childhood disease period (JDM), 90% of the patients were female in adult DM. Prior studies for adult DM referred to nearly similar results where male to female ratio was approximately 1 to 2.7.[15,16] Whereas, in JDM, the females are affected around twice as frequently as males.[17]

The current report exhibited distinctive skin photosensitivity of sun-exposed parts particularly heliotrope rash of the face and sometimes the upper limbs, sparing hidden areas, periorbital edema with heliotrope-rash, mask-like or butterfly-like facial violate scaly papules and macules, and Gottron papules/ sign. The characteristic cutaneous manifestations of DM strongly suggest that ultraviolet light plays

a significant role in the disease pathogenesis, and may modify the varying clinical and immunological appearance. However, the nature and the occurrence of photosensitivity in DM have been relatively poorly documented.[18-20] Previously, an abnormal cutaneous reaction to sunlight and ultraviolet radiation was reported, as 7 of 19 patients with DM had sunlight-induced worsening of their rash.[20] Abdel-Nasser *et al.* experienced the induction of ADM in a patient managed with ultraviolet light as a therapy for atopic dermatitis.[21]

The role and pathophysiology of photosensitive autoimmune reactions could be elucidated through ultraviolet-encouraged creation of tumor necrosis factor-a secretion that leads to apoptosis of keratinocyte and cellular antigens translocation, which then triggers the immune system.[22,23] The number of apoptotic keratinocytes in disordered basal zone junction of skin in DM was increased significantly, as compared with normal skin as confirmed by immunohistochemistry.[24,25]

The present data showed that all participants in both ADM and JDM complained of photosensitive reactions. ADM showed erythematous scaly papules and macules of the V-neck in 30% of subjects plus other findings as JDM. However, JDM revealed a high frequency of heliotrope rash with periorbital edema, facial violate scaly papules and macules- face mask/ butterfly distribution in 92%, and 77% of cases respectively, and surprisingly only 15.4% of patients experienced the Gottron papules/ sign. These results are higher than a previous study where heliotrope rash was seen in 49% and malar/ facial erythema in 49%, while Gottron's papules/ sign affected 53% of cases.[26]

A recent study found that 77% of cases with DM cases had periungual fold changes like capillary dilation, cuticular hemorrhage and hyperkeratosis.[27] While the present work showed shocking disagreement, where periungual erythema and telangiectasias were reported only in one (7.7%) adult patient. This difference could not well be explained but we speculate that it could be attributed to genetic and environmental changes as observed in many other clinical situations.

Another important clinical finding exhibited by patients in the present work was calcinosis cutis, which was experienced by 61.5% of JDM individuals. It was present on the upper limbs, lower extremities, and buttocks in 92%, 69.2%, and 30.8% of the patients, respectively. It is characterized by multiple hard tender nodules and sheetlike subcutaneous plaques. In contrast, this calcification was discovered in only 1 (10%) crippled adult woman. Valenzuela *et al.*[28] reported calcinosis in 31.3% patients. Other researchers reported that calcinosis cutis is more common in juvenile DM than in ADM patients, and the dystrophic deposition of calcium salts in the skin and soft tissues is a well-recognized distinct feature of juvenile DM, occurring in about 40% of all patients.[12,13,29] However, it is only around half as common in ADM, with an occurrence of around 20% and tends to arise later in the course of the disease.[28]

Another previous adult cohort study indicated the frequency of calcinosis to be 5.6%, and occurring at younger age while dysphagia and skin ulcer were independent of the development of calcinosis.[30]

Dystrophic calcinosis cutis refers to the precipitation of calcium in tissue proceeded by injury in patients with normal serum phosphorus and calcium. It is an uncommon condition associated with DM.[11] It has been assumed that local tissue damage and inflammation lead to change in the physiological mechanisms that normally prevent calcification. Then calcium

favorably deposits at an anatomical part where tissue damage occurs, including preceding panniculitis, lipodystrophy, discoid lesions, or trauma. These lesions may restore with scaring and secondary calcification. This calcification most frequently occurs on the shoulders, thighs, arms, breasts, and buttocks.[31] The lesions of dystrophic calcinosis cutis might have discomfort and pain, if it associated with panniculitis, ulceration, and secondary infection. These findings might indicate active DM.[12]

The current study has shown that 46.2% of affected children had overlying pyoderma and ulceration involving several locations such as the thigh, knees, elbows, hands and digital pulps. The possible explanations for ulcer development are as a consequence of repeated mechanical trauma, calcinosis, infection, and immune-mediated vascular damage with tissue necrosis. The existing outcomes show that whenever there was pyoderma, ulceration and scar in DM, there was underlying calcinosis and calcification. These finding were in contrast to previous studies where only the adults patients were affected by ulceration and chiefly when linked with anti-melanoma differentiation-associated gene 5 product (MDA5) antibody.[32,33] Several European cohorts described a significant relationship between ulceration and malignancy.[34,35] However, the present study showed no malignancy during the course of the disease.

Mechanic's hands are defined as a calcified hyperkeratotic rash on the thumb and the index finger, with desquamated papules, similar presentations to manual workers (eczema-like lesions) but have symmetrical lesions unrelated to the dominant hand and without associated pruritus and vesicles. In the existing work, Mechanic's hands were observed in 33.1% of patients with DM and was twice as common in children than adults. Findings are consistent with what was displayed in previous reports, where only 33% of cases with DM had mechanic's hands, and was considered as a valuable sign for diagnosis in individuals with lung diseases of collagen vascular disorders.[36,37]

The present work, fortunately, showed no malignancy in both JDM and ADM patients during the course of the disease. However, proceeding studies found that DM was linked with the increased danger of malignancy, and the risk was existing in all age groups and both genders and is highest after diagnosis within the first year.[29,38] The authors advise that adult patients of DM should be evaluated frequently for malignancy. We need further evaluation and investigation to explore this great diversity in the incidence of malignancy among patients with DM.

Conclusion

Twenty-three patients were evaluated. The presenting feature in the juvenile group was photosensitivity and a heliotrope rash of the face and calcinosis cutis. Photosensitivity with heliotrope dermatitis of the face and muscle weakness were observed in adults, with only one (10%) patient with calcinosis cutis (which was a common finding in patients with JDM). No malignancy was detected during follow-up in any patient. Photosensitivity is a common sign in patients with DM hence it must be considered as one of the major criteria, and should be included in the differential diagnosis of any patient with photo-distributed scaly erythema involving sun-exposed areas of the skin. Further study is needed in corporation with other related specialties to have more cases and more sophisticated clinical findings.

Declaration of patient consent The authors certify that they have obtained all appropriate patient consent.

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Conflict of interest Authors declared no conflict of interest.

Authors' contribution

KES, TAK, IKS Study design, acquisition of data, manuscript writing, analysis and interpretation of data, final approval of the version to be published.

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