A Delphi consensus on the nomenclature and diagnosis of lichen planus pigmentosus and related entities

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Abstract

Background: Although well known in clinical practice, research in lichen planus pigmentosus and related dermal pigmentary diseases is restricted due to lack of consensus on nomenclature and disease definition.

Aims and Objectives: Delphi exercise to define and categorise acquired dermal pigmentary diseases.

Methods: Core areas were identified including disease definition, etiopathogenesis, risk factors, clinical features, diagnostic methods, treatment modalities and outcome measures. The Delphi exercise was conducted in three rounds.

Results: Sixteen researchers representing 12 different universities across India and Australia agreed to be part of this Delphi exercise. At the end of three rounds, a consensus of >80% was reached on usage of the umbrella term 'acquired dermal macular hyperpigmentation'. It was agreed that there were minimal differences, if any, among the disorders previously defined as ashy dermatosis, erythema dyschromicum perstans, Riehl's melanosis and pigmented contact dermatitis. It was also agreed that lichen planus pigmentosus, erythema dyschromicum perstans and ashy dermatosis did not differ significantly apart from the sites of involvement, as historically described in the literature. Exposure to hair colours, sunlight and cosmetics was associated with these disorders in a significant proportion of patients. Participants agreed that both histopathology and dermatoscopy could diagnose dermal pigmentation characteristic of acquired dermal macular hyperpigmentation but could not differentiate the individual entities of ashy dermatosis, erythema dyschromicum perstans, Riehl's melanosis, lichen planus pigmentosus and pigmented contact dermatitis.

Limitations: A wider consensus involving representatives from East Asian, European and Latin American countries is required.

Conclusion: Acquired dermal macular hyperpigmentation could be an appropriate conglomerate terminology for acquired dermatoses characterised by idiopathic or multifactorial non-inflammatory macular dermal hyperpigmentation.

Key words: Acquired dermal macular hyperpigmentation, ashy dermatosis, consensus, erythema dyschromia perstans, lichen planus pigmentosus

How to cite this article: Sarkar R, Vinay K, Bishnoi A, Poojary S, Gupta M, Kumaran MS, *et al*. A Delphi consensus on the nomenclature and diagnosis of lichen planus pigmentosus and related entities. Indian J Dermatol Venereol Leprol 2023;89:41-6.

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Received: August, 2021 **Accepted:** November, 2021 **EPub Ahead of Print:** April, 2022 **Published:** January, 2023

DOI: 10.25259/IJDVL_804_2021 **PMID:** 35593293

Plain Language Summary

There is confusion in the distinction, clinical features and naming of skin diseases presenting with slate blue pigmentation. We undertook this exercise, wherein a series of questions were presented to a group of national and international experts in the field of pigmentary diseases. The questions were administered in rounds, with the results of the previous one being made known to all participants. A consensus of 80% was aimed at. At the end of three rounds, more than 80% agreed that the term 'acquired dermal macular hyperpigmentation' could be used to encompass all of the disorders and that there were minimal differences, if any, among them. The participants agreed that though both skin biopsy and dermatoscopy could diagnose the group as a whole, findings were not specific enough to differentiate the various entities included in the umbrella term.

Introduction

Hyperpigmentary dermatoses are commonly encountered by dermatologists and physicians in practice and are responsible for considerable morbidity and psychological distress, especially in dark skinned patients.[1] Acquired hyperpigmentary disorders can be broadly classified into predominantly *epidermal* hyperpigmentary diseases,[2] such as melasma, freckles, lentigines and post-inflammatory hyperpigmentation among others; and predominantly *dermal* hyperpigmentary diseases such as lichen planus pigmentosus (the prototype dermal pigmentary disease), Riehl's melanosis, ashy dermatosis, erythema dyschromicum perstans and pigmented contact dermatitis.[3] Although well known in clinical practice, research in acquired dermal pigmentary diseases is restricted due to lack of consensus on nomenclature, disease definition and clinical criteria.[4-6] These disorders have considerable clinico-pathological overlap, are difficult to treat and adversely affects patient's quality of life.

A recent global consensus coined the term 'acquired macular pigmentation of uncertain aetiology' as a unifying terminology for various morphologies of acquired dermal pigmentation. This was the first global attempt to define the clinical conditions lichen planus pigmentosus, ashy dermatosis, erythema dyschromicum perstans, Riehl's melanosis, pigmented contact dermatitis and idiopathic eruptive macular pigmentation.[3] The global consensus acknowledged the difficulty of categorising all patients into watertight compartments and encouraged classifying overlapping/undefined morphological entities into acquired macular pigmentation of uncertain aetiology. Herein, building on this global consensus statement, we attempted to further define and categorise acquired dermal pigmentary diseases in order to facilitate future research and patient management.

Delphi technique

The Delphi technique is a method of consensus building using a series of questionnaires to a panel of selected experts and stakeholders; the repetitive nature of this process, together with anonymous feedback by experts at each stage, allow convergence towards a consensus.[7,8] In this method, all participants get an equal opportunity to express their views, avoiding domination by a select group of experts. Furthermore, the results and responses of previous rounds are re-circulated among participants who are encouraged to reconsider their answers in the light of responses from other members.

Need for Delphi exercise

Recognising the grey areas and uncertainties surrounding acquired dermal hyperpigmentary diseases, informal discussions were carried out among Indian researchers interested in pigmentary diseases. For better defining these disease entities, core areas were identified including disease definition, etiopathogenesis, risk factors, morphology and distribution, diagnostic methods such as histopathology and dermatoscopy, treatment modalities and treatment outcomes. The Delphi exercise was undertaken by the special interest group on pigmentary disorders of the Indian Association of Dermatologists, Venereologists and Leprologists (IADVL) under the aegis of the IADVL Academy.

Materials and Methods

Panel selection

The group identified national and international researchers working on dermal hyperpigmentary diseases by a literature search in January 2020. A formal request to be part of the Delphi panel was mailed to first and corresponding authors of publications related to acquired dermal pigmentary diseases, published in high impact factor journals.

Delphi method

First round

In the first round, a Google form (Google Inc. California, United States of America) with 30 questions focusing on previously identified core areas and literature search were drafted and circulated among the Delphi members aiming at 80% consensus. The questions had multiple-choice answers with an option to express individual opinions if the members disagreed or had a viewpoint that was not listed. No in-person meetings were conducted because of restrictions during the period 2020-21.

Second and third round

If consensus could not be reached on any statement(s), new revised versions were circulated to participants in subsequent rounds 2 and 3.

Results

Participants

Sixteen researchers representing 12 different universities across India and Australia agreed to be part of this Delphi

exercise during round 1, and 14 and 11 responded in round 2 and 3, respectively. Among the respondents, 4 (25.0%) and 6 (37.5%) participants were seeing 7–10 and >10 new patients with these disorders per month.

Round one

After first round of the Delphi exercise, ≥80% agreement was reached for 12 of 30 questions, summarised in Table 1.

More than 75% agreement was reached at using the collective term 'acquired dermal macular hyperpigmentation' for lichen planus pigmentosus, pigmented contact dermatitis, Riehl's melanosis, erythema dyschromicum perstans and ashy dermatosis; and 12 (75%) members agreed that a combination of genetic predisposition, autoimmune diatheses and lichenoid tissue reaction to a variety of allergens played a role in their development.

An association with frontal fibrosing alopecia, endocrine disorders (especially hypothyroidism), vitiligo and atopy had been observed by eleven (68.8%), seven (43.5%), six (37.5%) and four (25%) members, respectively. Further, ten (62.5%) members observed the rare occurrence of mild powdery or furfuraceous scaling and six (37.5%) felt that erythema and scaling coincided in a few patients. Topical corticosteroids, topical calcineurin inhibitors, oral steroid sparing immunosuppressants or immunomodulators, oral mini-pulse of corticosteroids and lasers were being used routinely to treat these disorders by eleven (68.8%), four (25.1%), one (6.3%), zero (0%) and zero (0%) participants. Steroid sparing immunosuppressants or immunomodulators that were routinely used included isotretinoin, dapsone and colchicine by ten (62.5%), one (6.3%) and zero (0%) of respondents, respectively, whereas four (25%) used acitretin, azathioprine, cyclosporine, methotrexate and mycophenolate mofetil.

Table 1: Summary of statements on which ≥80% agreement was reached after round 1 of Delphi consultations

The incidence of acquired dermal hyperpigmentation is increasing.
There is a female predominance.
The most common age group affected is the middle aged (30–50 years).
Although classically described as non-inflammatory, the rare occurrence of mild lesional and perilesional erythema in patients of acquired dermal pigmentation does not negate the diagnosis.
Although traditionally described, the presence of the classical erythematous border is not essential to arrive at a diagnosis of erythema dyschromicum perstans. It was not seen in almost half of the patients who were given this diagnosis in practice of the Delphi members.
Lichen planus has an association with acquired dermal pigmentation, especially the lichen planus pigmentosus type.
There seems to be an association with sun-exposure and usage of cosmetics.
Skin biopsy is routinely performed while managing these patients.
Patch testing and photo-patch testing are useful, especially in patients with clinically or historically apparent photo-aggravation.
Factors that affect the management of acquired dermal pigmentation in practice include: activity, extent of involvement, sites of involvement, degree of hyperpigmentation and duration of disease.

Round two

Fourteen participants responded to the second round of Delphi, more than 80% of participants felt at this stage that the dermatoses described previously as lichen planus pigmentosus, pigmented contact dermatitis, Riehl's melanosis, erythema dyschromicum perstans and ashy dermatosis were interrelated/overlapping disorders and should be clubbed together. They agreed with the usage of the umbrella term 'acquired dermal macular hyperpigmentation' for all these dermatoses, with 10 (71.4%) agreeing with a further classification of acquired dermal macular hyperpigmentation under two broad headings – *acquired dermal macular hyperpigmentation with contact sensitisation* and *acquired dermal macular hyperpigmentation without contact sensitisation.*

Importantly, ≥80% participants agreed that apart from the presence of an erythematous border, erythema dyschromicum perstans did not differ significantly from ashy dermatosis; and that pigmented contact dermatitis and Riehl's melanosis were similar entities, with 10 (71.4%) respondents agreeing that lichen planus pigmentosus, erythema dyschromicum perstans and ashy dermatosis did not differ significantly apart from the sites of involvement described historically in the literature. Both lichen planus pigmentosus and pigmented contact dermatitis were thought to be photosensitive disorders by seven (50.0%) of the participants and only eight (57.1%) members felt that idiopathic eruptive macular pigmentation should be included in the spectrum of acquired dermal macular hyperpigmentation.

At this stage, ≥80% members agreed that histopathology could not reliably differentiate these disorders with ten (71.4%) agreeing that band-like lichenoid infiltrate was not specific for a diagnosis of lichen planus pigmentosus. Further, ten (71.4%) felt that histopathology was both diagnostic and prognostic for the entity of acquired dermal macular hyperpigmentation, with nine (64.2%) agreeing that inflammatory changes on histopathology could be used to assess the disease activity of these disorders, signifying an active stage and formed a basis for starting systemic immunosuppressants/immunomodulators.

Regarding dermatoscopy, 11 (78.6%) felt that it could help in diagnosing these dermatoses in an appropriate clinical setting by demonstrating dermal pigmentation and could obviate the need of skin biopsy, with ten (71.4%) agreeing that the role of dermatoscopy in the management of these disorders was both diagnostic and prognostic. However, ten (71.4%) agreed that dermatoscopy, too could not reliably differentiate the individual disorders clubbed under acquired dermal macular hyperpigmentation.

Majority (≥80%) consensus was reached about the requirement for an objective severity scoring scale for assessing therapeutic response in these dermatoses, with

eight (57.1%) agreeing that dermal pigmentation and area severity index (described previously for scoring severity of these dermatoses when affecting face and neck) could be used for research and six (42.9%) agreeing that it could be used in both research and clinical settings [Table 2].

Round three

The final round of the Delphi exercise, was completed with eleven participants responding and ≥80% respondents now agreed that lichen planus pigmentosus, erythema dyschromicum perstans and ashy dermatosis did not differ significantly apart from the sites of involvement described historically in the literature and ≥80% consensus was again reiterated on the inclusion of lichen planus pigmentosus, pigmented contact dermatitis, Riehl's melanosis, erythema dyschromicum perstans and ashy dermatosis under the broad heading of acquired dermal macular hyperpigmentation with further classification as acquired dermal macular hyperpigmentation with and without contact sensitisation; as well as diagnostic and prognostic role of dermatoscopy in these disorders. Table 2 summarises the statements where ≥80%, ≥70–80% and <70% consensus was reached after the completion of Delphi Rounds 2 and 3.

At this stage, the participants were also asked to fill in a table [Table 3] to summarise their perception of clinico-demographic features of lichen planus pigmentosus, pigmented contact dermatitis, Riehl's melanosis, erythema dyschromicum perstans and ashy dermatosis.

Table 2: Summary of statements on which ≥80%, ≥70–80% and <70% consensus was reached after the completion of Delphi Rounds 2 and 3

S. No.	Statements where ≥80% agreement was achieved amongst the Delphi members
1.	Apart from the presence of an erythematous border, erythema dyschromicum perstans does not differ significantly from ashy dermatosis.
2.	Riehl's melanosis and pigmented contact dermatitis are similar entities.
3.	Lichen planus pigmentosus, erythema dyschromicum perstans and ashy dermatosis do not differ significantly apart from the sites of involvement described historically in literature.
4.	There seems to be an association with the sun-exposure and usage of cosmetics, especially hair colours.
5.	Dermatoses described previously as lichen planus pigmentosus, pigmented contact dermatitis, Riehl's melanosis, erythema dyschromicum perstans and ashy dermatosis are interrelated/overlapping disorders and could be clubbed together.
6.	'Acquired dermal macular hyperpigmentation' could be a suitable umbrella term for dermatoses described previously as lichen planus pigmentosus, pigmented contact dermatitis, Riehl's melanosis, erythema dyschromicum perstans and ashy dermatosis.
7.	Dermatoscopy could help in diagnosing acquired dermal macular hyperpigmentation in an appropriate clinical setting by demonstrating dermal pigmentation and could obviate the need of skin biopsy.
8.	There is a requirement for the usage of an objective severity scoring scale/dermal pigmentation and area severity index for assessing therapeutic response in these dermatoses.
S. No.	**Statements where ≥70–80% agreement was achieved amongst the Delphi members**
1.	Acquired dermal macular hyperpigmentation could be further classified under 2 broad headings – *acquired dermal macular hyperpigmentation with contact sensitisation* and *acquired dermal macular hyperpigmentation without contact sensitisation.*
2.	Band-like lichenoid infiltrate was not specific for a diagnosis of lichen planus pigmentosus.
3.	Histopathology was both diagnostic and prognostic for acquired dermal macular hyperpigmentation.
4.	Dermatoscopy was both diagnostic and prognostic for acquired dermal macular hyperpigmentation.
5.	Dermatoscopy and histopathology could not reliably differentiate the individual disorders clubbed under acquired dermal macular hyperpigmentation.
6.	A combination of genetic predisposition, autoimmune diatheses and lichenoid tissue reaction to a variety of allergens played a role in the development of acquired dermal macular hyperpigmentation.
S. No.	**Statements where <70% agreement was achieved among the Delphi members**
1.	Idiopathic eruptive macular pigmentation should be included in the spectrum of acquired dermal macular hyperpigmentation (57% of participants agreed).
2.	Both lichen planus pigmentosus and pigmented contact dermatitis were thought to be photosensitive (50% of participants agreed).
3.	Dermal pigmentation and area severity index (described previously for scoring severity of these dermatoses when affecting face and neck) could be used for research (57% of participants agreed) and in both research and clinical settings (43% of participants agreed).
4.	Rarely, mild powdery or furfuraceous scaling occurs with lesions of acquired dermal macular hyperpigmentation (62% of participants agreed) and erythema and scaling coincided in a few patients (35% of participants agreed).
5.	Inflammatory changes on histopathology could be used to assess the disease activity of these disorders and formed a basis for starting systemic treatment (64% of participants agreed).
6.	An association with frontal fibrosing alopecia, endocrine disorders (especially hypothyroidism), vitiligo and atopy had been observed (70%, 41%, 35% and 23% of participants agreed, respectively).
7.	Topical corticosteroids, topical calcineurin inhibitors, oral steroid sparing immunosuppressants or immunomodulators, oral mini-pulse of corticosteroids and lasers were being used routinely to treat these disorders (69%, 23%, 8%, 0% and 0% of participants agreed, respectively).
8.	Steroid sparing immunosuppressants or immunomodulators that were routinely used included isotretinoin, dapsone and colchicine (64%, 7% and 0% of participants agreed, respectively). Other agents that were used included acitretin, azathioprine, cyclosporine, methotrexate and mycophenolate mofetil (28% of participants agreed).

Table 3: Summary of the perception of the participants regarding the clinicodemographic features of lichen planus pigmentosus, pigmented contact dermatitis, Riehl's melanosis, erythema dyschromicum perstans and ashy dermatosis

	Age (range, years)	Age (commonest, years)	Gender	Lichen planus associated	Sites	Precipitating factor, pruritus
Lichen planus pigmentosus	12–60	30–50	Females	Yes	Face, neck, trunk, upper limbs and flexures	Yes, yes
Riehl's melanosis	20–60	30–50	Female	No	Forehead, zygomatic area, neck and lateral cheeks	Yes (perfumes, hair colours), yes
Pigmented contact dermatitis	20–60	30–50	Female	No	Forehead, zygomatic area, neck and lateral cheeks	Yes (perfumes, hair colours), yes
Erythema dyschromicum perstans	7–65	10–20	Male	No	Trunk	Absent, absent
Ashy dermatosis	20–65	30–50	Female	No	Trunk, upper limbs	Absent, absent

Discussion

Acquired dermal macular hyperpigmentation is a recently introduced terminology with an aim to unify the disorders previously known by the names of ashy dermatosis, erythema dyschromicum perstans, Riehl's melanosis, pigmented contact dermatitis and lichen planus pigmentosus.[3,4,9,10] The rationale for this approach is the significant clinico-pathologic similarities between the individual entities along with their rarity and the benefits of a unified terminology in patient management and communication among researchers at a global level. The term acquired dermal macular hyperpigmentation provides information about the origin and natural history (acquired), localisation (dermal) and character (macular) of the hyperpigmentation; and seems to be a more inclusive terminology. In the current Delphi exercise, dermatologists actively involved in the care of patients presenting with this group of disorders agreed with the use of the term acquired dermal macular hyperpigmentation.

Majority of participants agreed with the statement that the absence of an erythematous border did not deter one from making a diagnosis of erythema dyschromicum perstans and apart from this erythematous border, there was no appreciable difference between erythema dyschromicum perstans and ashy dermatosis.[11-13] The participants also agreed that erythema may be occasionally seen in patients having a disease phenotype that was closer to lichen planus pigmentosus/pigmented contact dermatitis rather than erythema dyschromicum perstans, as classically described.

Participants also agreed that ashy dermatosis, erythema dyschromicum perstans and lichen planus pigmentosus presented with morphologically similar lesions, with the only difference being their distribution. Similarly, Riehl's melanosis and pigmented contact dermatitis were agreed to be almost similar entities with no appreciable differences.

Participants agreed that skin biopsy was routinely performed in the management of these patients, but the aim of biopsy was not to differentiate between individual entities. Similarly, participants agreed that dermatoscopy was routinely performed but not in order to differentiate the entities. Its main importance was to identify features of dermal pigmentation characterised by dots and globules, thereby obviating a need for skin biopsy in clinical settings with high pre-dermatoscopy probability of acquired dermal macular hyperpigmentation.[10,14]

Overall, it was agreed that the individual entities of ashy dermatosis, erythema dyschromicum perstans, Riehl's melanosis, pigmented contact dermatitis and lichen planus pigmentosus had significant clinico-pathologic similarities. Participants agreed that undertaking patch tests in patients with acquired dermal macular pigmentation could identify the subset that probably developed their pigmentation secondary to non-eczematous delayed lichenoid dermatitis type-IV hypersensitivity to certain antigens, specifically hair colours and cosmetics. Similarly, it was agreed that a better classification of acquired dermal macular hyperpigmentation would be that into the broad headings of '*with and without contact sensitisation'* since it would help with the management of patients if such antigens could be identified and eliminated.[9,14]

There were certain statements where a consensus could not be achieved. Only 57% of participants felt that idiopathic eruptive macular pigmentation can be included under acquired dermal macular hyperpigmentation.[15] Similarly, 70% consensus was achieved regarding the association of these disorders with lichen planus and frontal fibrosing alopecia respectively. The association with atopic tendency, thyroid disorders and vitiligo did not reach much agreement.[16-20] Both Riehl's melanosis/pigmented contact dermatitis and lichen planus pigmentosus were considered to be equally photosensitive by 50% of the participants.

Limitations

The aim of this exercise was to reach at a consensus regarding the salient features relating to acquired dermal macular hyperpigmentation and included dermatologists from Indian subcontinent and Australasian continent. It could have been more inclusive of the experts from other geographical domains; however, it was not deemed feasible due to operational reasons.

Conclusion

To conclude, this Delphi consensus agreed that acquired dermal macular hyperpigmentation could be an appropriate

conglomerate terminology for dermatoses characterised by idiopathic or multifactorial non-inflammatory macular dermal hyperpigmentation. This shall hopefully bring about a much-needed uniformity with a simple sub-classification suggested by us, when further research regarding the pathogenesis, clinico-pathology, dermatoscopy and treatment of these rare dermatoses is reported. This can also help in treating and prognosticating patients by dermatologists both in research institutes and in practice.

Declaration of patient consent

Patients' consent not required as there are no patients in this study.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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