

# Perinevoid Alopecia: Report of Two Cases and Literature Review

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**Abstract:** Perinevoid alopecia (PA) is a rare variant of alopecia areata (AA) associated with a central pigmented nevus. In this study, we reported two cases of PA and reviewed 14 cases from 11 studies in the literature. In one of our cases, PA was combined with a halo nevus and white terminal hairs were spared in the hair loss patch, which was rarely reported in the literature. It is implicated that antigens from melanocytes might be involved in the development of AA in PA.

**Keywords:** alopecia areata, hair loss, nevus, halo nevus

## Introduction

Perinevoid alopecia (PA), first described by Yesudian and Thambiah in 1976, is a variant of alopecia areata (AA) associated with a central pigmented nevus.<sup>1,2</sup> We found 14 cases from 11 studies in the literature from 1976 to 2022. PA is an extremely rare disorder with a clinically distinctive feature of alopecic patch with a central pigmented nevus and a histologically specific finding of inflammatory cell infiltration in nevus cell nests and perifollicular areas.<sup>4</sup> With dermoscopy, the manifestations of both central nevi and AA were revealed. Although the pathogenesis is still unclear, it is thought that PA is secondary to an inflammatory response against nevus cells or melanocytic structures.<sup>1</sup> We reported the clinical findings of two patients with PA.

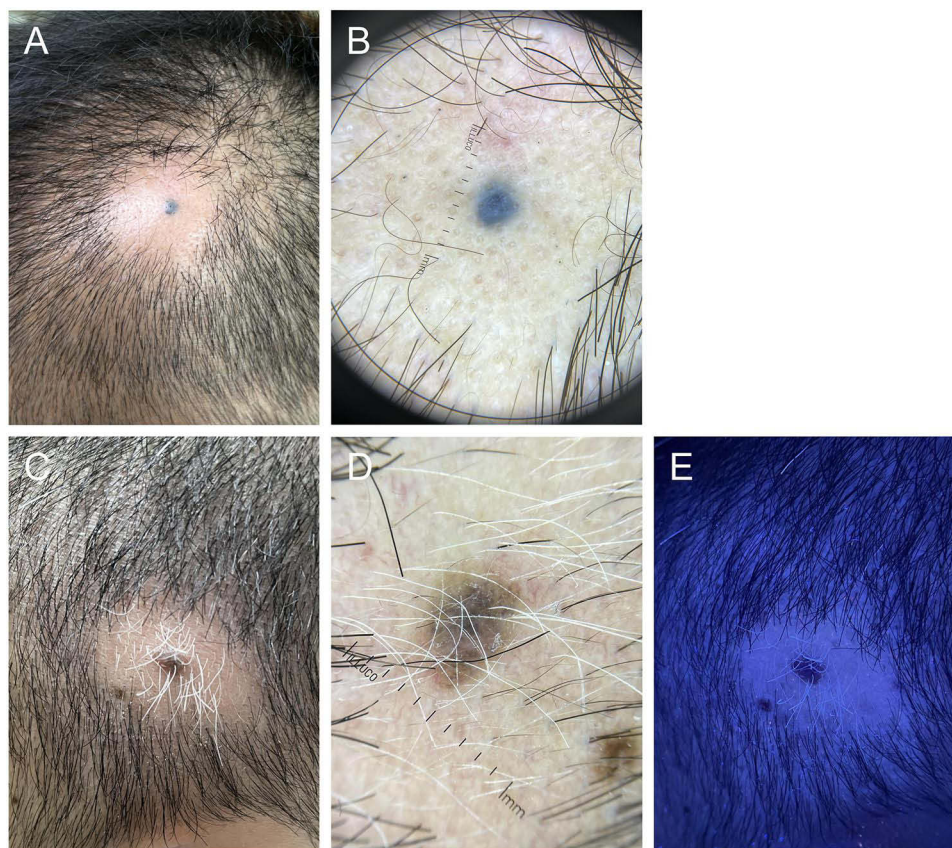
## Case Reports

### Case 1

A 46-year-old Chinese male complained of a patchy hair loss with a central blue nevus on his left temporal (Figure 1A). The nevus had been presented more than ten years, while the patch of hair loss was noticed one month prior to the clinic visit. Upon dermatological examination, an area of nonscarring alopecia, approximately 2.0×2.0 cm, without signs of inflammation, surrounding a 4-mm diameter nevus, was observed in the left temporal region. Dermoscopy examination revealed a well-circumscribed lesion with homogenous blue-grayish globular pattern, broken hairs, black dots, yellow dots and short vellus hairs (Figure 1B). The diagnosis of PA was made based on the clinical findings, and two sessions of intralesional steroid injections with a month interval resulted in a complete recovery of hair growth.

### Case 2

A 28-year-old Chinese male patient began to experience a patchy hair loss with a central nevus and surrounding hypopigmentation in the left temporal region two months ago, which increased in size progressively (Figure 1C). The central pigmented nevus had been presented more than ten years, while the asymptomatic patch of hair loss was noticed two months prior to the clinic visit. Dermatological examination revealed a 2-mm diameter dark brown pigmented papule, surrounded by a 0.7-cm diameter area of non-scarring alopecia, and white terminal hairs in the area of previous hair loss were unaffected. With dermoscopy, we noticed a globular, dark brown pigmented tumor with peripheral areas of



**Figure 1** Case 1: (A) Macroscopic image showing an area of nonscarring alopecia, approximately 2.0×2.0 cm, without signs of inflammation, surrounding a 4-mm diameter well-circumscribed nevus, was observed in the left temporal region. (B) Dermoscopic image showed a well-circumscribed lesion with homogenous blue-grayish globular pattern, broken hairs, black dots, yellow dots and short vellus hairs. Case 2: (C) Macroscopic image revealed a patchy hair loss with a central nevus and surrounding hypopigmentation in the left temporal region. (D) With dermoscopy, we noticed a globular, dark brown pigmented tumor with peripheral areas of light-brown pigment, short vellus hairs and broken hairs. (E) Wood's light examination of the area of alopecia was positive with fluorescence enhancement.

light-brown pigment, short vellus hairs and broken hairs (Figure 1D). Wood's light examination of the area of alopecia was positive with fluorescence enhancement (Figure 1E). Halo nevi (HN) consists of central pigmented nevi surrounded by a sharp zone of hypopigmentation, so the diagnosis of PA and HN was made. He was treated with topical minoxidil (5% solution) while complete hair regrowth was observed after a few months.

## Discussion

We preferred to define PA as alopecia areata around a pigmented nevus and summarized some clinical findings.<sup>3</sup> Various pigmented nevi in 14 cases from 11 studies were found (Table 1), including intradermal nevi (n=5), compound nevi (n=2), blue nevi (n=3), etc.<sup>1-11</sup> A blue nevus and a melanocytic nevus were observed in our cases. It seems PA may affect more males than females. In all the 16 reported cases, 13 were males and 3 were females. The age of these patients was from 19 to 46 years old, and the average age was 31.06 years old. The hair loss patches were most frequently located on the vertex (n=7) and occipital (n=5), followed by the beard area (n=1), the back of the scalp (n=1), chin (n=1) and neck area (n=1).

Dermoscopy examination revealed manifestations of both central nevi and AA. Dermoscopy is the noninvasive diagnostic equipment used increasingly in dermatological practice. In all 5 cases with dermoscopy alterations (including our two cases), yellow dots (n=3), vellus hairs (n=3), black dots (n=2), exclamation mark hairs (n=2), and broken hairs (n=2) were identified, which were consistent with active alopecia areata. A well-circumscribed lesion with homogenous blue-grayish globular pattern and a globular, dark brown pigmented tumor with peripheral areas of light-brown pigment were observed in our cases. Of note, one of our patients had a typical patch of AA with a central halo nevus and remaining white terminal hairs in the area of hair loss, which had not been reported previously.

**Table 1** 14 Cases of Perinevoid Alopecia Reported in the Literature

Author	Sex/ Age (Year)	The Features of Nevus	The Features of Alopecia	Dermoscopy Findings	Pathology	Diagnose	Treatment	Prognosis
Ruiz-Arriaga <sup>1</sup>	M/35	Dysplastic nevi, melanocytic nevus, a halo nevus	A patch of alopecia	Exclamation mark hairs, yellow dots, and vellus hairs	An intradermal melanocytic nevus. Inflammatory cell infiltration: nevus cell nests(+), hair follicle(+), perifollicular areas(+)	Perinevoid alopecia	Surgical excision	Hair regrowth
Yesudian <sup>2</sup>	M/29	Two nevi: 1. A pigmented, nevocellular nevus 2. A nevus	Two patches of baldness: 1. on the vertex 2. on the beard area in the right submandibular region	/	Type of nevus: a compound nevus. Inflammatory cell infiltration: hair follicle(+)	Perinevoid alopecia	Surgical excision	Hair regrowth
Yesudian <sup>2</sup>	M/20	A pigmented nevus	Several patches of baldness: 1. several patches in the occipital region 2. one patch shows central nevus	/	Type of nevus: a compound nevus; Inflammatory cell infiltration: hair follicle(+)	Perinevoid alopecia	Surgical excision	Hair regrowth
Yesudian <sup>2</sup>	M/29	A pigmented nevus on the chi	Three patches of baldness: 1. two patches on the scalp 2. a single patch on the chin	/	Type of nevus: an intradermal nevus	Perinevoid alopecia	Surgical excision	No hair regrowth for 1 month
Kimura <sup>3</sup>	F/22	Two melanocytic nevi	Two hairless patches: 1. on the right side of the vertex 2. on the back of the scalp	Yellow dots, black dots, exclamation mark hair, globules and typical pseudonetwork in the black nodule	Inflammatory cell infiltration: nevus cell nests(+), hair follicle(+)	Perinevoid alopecia	Surgical excision	Hair regrowth
Kim <sup>4</sup>	F/33	A skin-colored papule	A patch of alopecia: on the vertex	/	Inflammatory cell infiltration: nevus cell nests (+), perifollicular areas(+)	Perinevoid alopecia	Surgical excision	Hair regrowth
Heng <sup>5</sup>	M/20	A compound melanocytic naevus combined with a blue naevus	A patch of alopecia: from the vertex to midocciput	/	Type of nevus: a combined naevus	Perinevoid alopecia	Intralesional steroids	Follow up

(Continued)

Table 1 (Continued).

Author	Sex/ Age (Year)	The Features of Nevus	The Features of Alopecia	Dermoscopy Findings	Pathology	Diagnose	Treatment	Prognosis
Yan Hongbo <sup>6</sup>	M/37	A cellular blue nevus	A patch of alopecia: on the left vertex; the length of regrowth hair was about 1 cm	/	Type of nevus: a intradermal naevus	Cellular blue nevus; intradermal nevus; alopecia	No treatment	Follow up
Li Lina <sup>7</sup>	F/20	Two nevi: 1. A melanocytic nevus 2. A dark red neoplasm	Two patches of baldness: 1. on the occipital scalp for 2 months 2. on the neck area	/	Type of nevus: a intradermal naevus; Inflammatory cell infiltration: hair follicle(+)	Intradermal nevi; alopecia areata; perinevoid alopecia	Excision	Hair regrowth
Ma Xiaolei <sup>8</sup>	M/38	A congenital pigmented nevus	A patch of alopecia: on the scalp; delicate hair regrowth	/	Type of nevus: a intradermal naevus. Inflammatory cell infiltration: hair follicle(+)	Congenital pigmented nevus; alopecia areata	Intralesional steroid injections, topical corticosteroids, etc	Follow up
Machado <sup>9</sup>	M/19	A pigmented nevus	A patch of alopecia: on the occiput	Compatible with alopecia areata	/	Perinevoid alopecia	Unknown	/
Pu Xiongming <sup>10</sup>	M/32	A pigmented nevus	A patch of alopecia: on the occipital scalp	/	/	Alopecia areata	Excision; intralesional steroid injections; hair tonic	Hair regrowth
Pu Xiongming <sup>10</sup>	M/46	A pigmented nevus	A patch of alopecia: on the right side of the vertex	/	/	Alopecia areata	Excision; intralesional steroid injections; hair tonic	No hair regrowth
Takeichi <sup>11</sup>	M/43	A blue-gray macule with sparse hairs on the right vertex	Two patches of baldness: 1. on the right vertex 2. on the left temporal region	/	Type of nevus: blue nevus, cellular type; Inflammatory cell infiltration: hair follicle(+)	Alopecia areata	Topical application of corticosteroid for 3 months	Complete recovery of hair growth

Histopathologically, inflammatory cell infiltration was most frequently located in perifollicular areas. Skin biopsy in 11 cases showed inflammatory cell infiltration in the hair follicle (HF) (n=7), nevus cell nests (n=3), and perifollicular areas (n=2). Nevus cell nests were present in the upper dermis almost in all cases.

The pathogenesis of PA is unclear, and it might be secondary to an inflammatory response against nevus cells or melanocytic structures.<sup>1</sup> HF-derived autoantigens (eg, keratinocyte and melanocyte antigen) have long been suspected as the potential specific autoantigen for AA.<sup>12</sup> PA, defined as AA around a pigmented nevus, is a special variant of AA and may be valuable for the better understanding of the pathogenesis of AA. In one of our cases, we found a patch of alopecia with a central halo nevus and white terminal hairs spared in the hair loss patch. A halo phenomenon around a nevus has been suggested to be due to immunologic responses against melanocytes.<sup>13</sup> White terminal hairs lack melanocytes so they are not attacked by CD8<sup>+</sup> T cells. What's more, in this case, Wood's light examination of the area of alopecia was positive with fluorescence enhancement, representing the loss of melanin, which indicated PA might be secondary to an inflammatory response against melanocytes. Unfortunately, no case about PA observed under Wood's lamp was found and objective biopsy was refused in this case. Moreover, hair regrowth was observed after surgical removal of the whole lesion of the nevus in 7 cases. Therefore, we postulated that the immune cells around the naevi led to an attack on the peripheral HF similar to the autoimmune destruction of melanocytes in a halo naevus. Some antigens of nevus cells were assumed to be similar to the antigens of anagen hair bulbs, such as antigens of melanocytes or melanin granules. Antigens from melanocytes might be involved in the development of AA in PA.

Diagnosis and treatment of PA were summarized. The diagnoses of this disease in 14 cases were PA (n=8), AA (n=5), nevi (n=3), and alopecia (n=1). PA might be misdiagnosed as AA, nevi or alopecia because of unawareness of this rare condition, and the central pigmented nevus was frequently overlooked. Therefore, it is worth reporting this case series to raise the awareness of this rare condition. In most cases, surgical removal of the whole lesion of the nevus (n=9) might lead to hair regrowth (n=7). However, there were two cases that showed no significant hair regrowth after excision of the central nevus. Intralesional steroids may also be effective, as in one of our cases.

A pigmented nevus may lead to inflammatory cell infiltration and immune response, causing peripheral vitiligo or AA. Why it causes these differences remains to be elucidated. Further researches are required to clarify the mechanisms of PA. This special variant of AA may be valuable for the better understanding of pathogenesis and further treatment development of AA.

## Abbreviations

PA, perinevoid alopecia; AA, alopecia areata; HN, halo nevus; HF, hair follicle.

## Consent Statement

Written informed consent was provided to the patient to have the case details and associated images published. Institutional approval was not required to publish the case details.

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## Disclosure

The authors declare no conflict of interest.

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