

## Case Report



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# Isolated Patchy Heterochromia of the Scalp Hair: A Rare Entity with Literature Review

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## ABSTRACT

Scalp hair heterochromia involves the presence of two different colors of the scalp hair in the same individual. It may be of three types: patchy, diffuse, and segmental. Isolated patchy heterochromia of the scalp hair is a rare entity, and a very few cases have been described in the literature. Hereby, we report one such case of isolated patchy scalp hair heterochromia in a 2-year-old healthy male child with black hairs presenting with a bunch of blond hairs without any underlying abnormalities along with the literature review.

**Key words:** Blond hairs, heterochromia, scalp

## INTRODUCTION

Heterochromia of the scalp hair is a rare phenomenon characterized by the presence of two distinct colors of the scalp hair in the same person.<sup>[1]</sup> Typically, colors such as brown, blond, red, and yellow hair presenting in the background of dark color hairs have been described in previous studies.<sup>[1-11]</sup> Here, we present a pediatric patient with scalp hair heterochromia without any underlying abnormalities.

## CASE REPORT

A 2-year-old male child of Indian origin with black hairs presented with a bunch of blond hairs of the same length on the right and left parietal region of the scalp since birth [Figure 1]. The patch of blond hairs approximately measured about 6 cm × 8 cm. The patient was in a good health with normal developmental milestones. Family and past history was also unremarkable. There was no history of exposure to any other chemicals, trauma, or inflammatory disease of the scalp.

General physical examination revealed normal physical development. On cutaneous examination, the patient had tuft of blond hair on the bilateral parietal region of the scalp following Blaschko's lines with normal color of the underlying skin. As the underlying skin of blond hairs was

normal, the diagnosis of melanocytic nevi, halo nevus, and vitiligo was excluded. The hair of the eyelashes and eyebrows were of black color. Nails, teeth, and oral mucosa were normal. No ophthalmologic, audiology, or neurological abnormalities were found. Routine blood investigations were normal. On light microscopy, the lighter hair appears to be slightly thinner than the darker hair and the pigmentation was homogenous along the entire hair shaft [Figure 2]. As the pigmentation was homogenous throughout the hair shaft, the diagnosis of segmental heterochromia was excluded. Based on these findings, diagnosis of patchy heterochromia of the scalp hair was made.

## DISCUSSION

Follicular melanogenesis of hairs is tightly coupled with hair growth cycle, i.e., melanocytic proliferation (during

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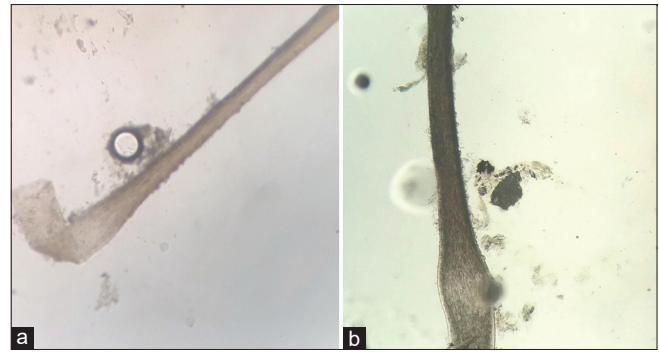


**Figure 1:** Tuft of blond hairs on the right and left parietal region with black hairs on the rest of the scalp in a 2-year-old male child

**Table 1: Etiology of Heterochromia of scalp hair<sup>3</sup>**

Genetic basis
Nutritional defects
Kwashiorkor (Flag sign)
Severe Iron deficiency anaemia
Vitamin B12 deficiency
Copper deficiency- Menke's syndrome
Metabolic defects
Phenylketonuria
Homocystinuria
Oasthouse disease
Drugs
(a) Minoxidil
(b) Diazoxide
(c) Chloroquine/hydroquinone
(d) Resorcin
(e) Dithranol and Chrysarobin stain
(f) Mephenesin
(g) Triparanol
(h) Furobutyrophenone
(i) Phenylthiourea
(j) Anti-Parkinson drugs- Bidopa, bromocriptine
(k) Oral etretinate therapy
5. Melanocytes nevi
6. Poliosis
(A) Hereditary defects
(a) Piebaldism
(b) Tietz syndrome
(c) Waardenburg syndrome
(d) Vogt-Koyanagi-Harada syndrome
(B) Acquired defects
(a) Inflammatory processes
(b) X- irradiation
(c) Dental treatment (beard area)
7. Vitiligo
8. Albinism
9. Chediak-Higashi Syndrome
10. Accidental causes
(a) Copper exposure- green hair
(b) Cobalt workers- white- blue hair
(c) Tar in cigarette smoke- yellow hair
(d) Trinitrotoluene- reddish-brown hair

early anagen), maturation (mid-to-late anagen), and melanocytic death via apoptosis (during early catagen). The process of melanogenesis is controlled by complex genetic mechanisms. Melanin pathway divides into two pathways after the formation of dopaquinone from



**Figure 2:** Under light microscopy, (a) showing blond hair and (b) showing black hair; blond hair is thinner than black hair and pigmentation is uniform in black hair as compared to blond hair (×10 magnification)

tyrosine. Dopaquinone converts to 5,6-dihydroxyindole and 5,6-dihydroxyindole-2-carboxylic acid oligomers to form eumelanin. Another pathway leads to the formation of benzothiazinyl alanine which forms pheomelanin and trichochromes. Tyrosinase activity determines the color of individual hair. The skin phototype does not determine the hair color as can be seen by the co-expression of eumelanin hair color in the Fitzpatrick skin type 1 and 2. Melanosome structure correlates with the type of melanin produced. Black hair follicle melanocytes have the largest number of eumelanosomes (melanosomes containing eumelanin) and red hair follicle melanocytes contain pheomelanosomes. Whereas, melanosomes in blond hair are weakly melanized. The author also speculates that hair follicles individually may have remarkable autonomy in terms of their pigment type, which may be the cause of variable polymorphisms in hair pigmentation.<sup>[12]</sup>

There are various etiologies of scalp hair heterochromia, which includes genetic basis, metabolic defects, nutritional defects, or drugs [Table 1].<sup>[3]</sup> The distribution of heterochromic hair can be symmetric or asymmetric. When the heterochromic hair is symmetric such as difference in scalp and body hairs, it is considered as physiologic. On the contrary, asymmetric distribution indicates an underlying pigmentary disorder.<sup>[1,2]</sup>

There are three different clinical types of asymmetrical scalp hair heterochromia: patchy, segmental, and diffuse type.<sup>[4]</sup> Patchy heterochromia of hairs which is not associated with any detectable skin pigmentary mosaicism, not characterized by poliosis and not resulting from the presence of an underlying melanocytic nevus, is known as isolated patchy heterochromia (IPH).<sup>[2]</sup>

Patchy and diffuse heterochromia are assumed to have genetic basis, whereas segmental heterochromia, which is characterized by alternating dark and light segments on each hair, is mostly associated with iron-deficiency anemia.<sup>[2]</sup> The review of literature of all the cases of patchy heterochromia of the scalp hair is given in Table 2.<sup>[1-11]</sup> In all these cases, the heterochromic hairs follow the lines of Blaschko. Our patient is a case of IPH along the lines of Blaschko, which might be secondary somatic mosaicism of one or more genes involved in pigmentation and skin pigmentation (hypo- or hyperpigmentation) due to the fact that such cutaneous mosaicism may not be present at birth but may develop later in life.<sup>[4]</sup>

There are only a few case reports of scalp hair heterochromia in the literature and none of them are from India. To the best of the authors' knowledge, ours might be the first case report of IPH of the scalp hair from India. This might

be due to the fact that this condition is asymptomatic and may be underdiagnosed so far. However, such IPH may be associated with other abnormalities of skin pigmentation. Further, genetic analysis of such pigmentary mosaicism is difficult as there is involvement of heterogeneous genes. Thus, dermatologist fraternity must be always aware of such possible pigmentary changes and keep these patients on regular follow-up.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parents have given their consent for the patient's images and other clinical information to be reported in the journal. The patient's parents understand that the patient's name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Table 2: Patchy heterochromia of hairs in the literature**

Authors	Year	Age (years)/sex	Onset	Race/ethnicity	Site/color of hair	Other associations
Restano <i>et al.</i> <sup>[2]</sup>	2001	Case 1-3/male	All since infancy	Not mentioned	Vertex and nuchal area/brown	None
		Case 2-5/male			Left parietal and occipital area/blond	None
		Case 3-35/male			Anterior to the whorl at vertex/blond	None
		Case 4-4/female			Occipital area/blond	None
		Case 5-10/female			Distributed diffusely/blond hair Case 1, 2, 3, and 5 followed the Blaschko's lines	Diffuse whorled bands of hypo/hyperpigmentation on skin, scoliosis, speech delay, and abnormal facies
Iorizzo <i>et al.</i> <sup>[3]</sup>	2007	Case 1-17/female	All congenital	Not mentioned	From midline along the right side of the scalp/ light brown color hair	None
		Case 2-6/male			Crown region/lighter hair	None
		Case 3-8/female			Vertex/brown color hair	None
		Case 4-7/male			Occipital area/light brown color hair	None
					All followed the Blaschko's lines	
Qiao and Fang <sup>[6]</sup>	2010	11/male	Congenital	Chinese origin	Vertex/brown color hair Consistent with Blaschko's lines	None
Zeng <i>et al.</i> <sup>[7]</sup>	2011	11/male	Since infancy	Chinese origin	Vertex/brown color Showed arrangement in Blaschko's lines on presented clinical picture	Lipematous scalp
Bonamonte <i>et al.</i> <sup>[8]</sup>	2014	5/male	Congenital	Italian origin	Numerous patches, especially over temporo-auricular and frontoparietal area	None
Park <i>et al.</i> <sup>[4]</sup>	2015	8 months/male	Congenital	Korean origin	Left parietal scalp/brown Vertical band shaped following the lines of Blaschko	Congenital bilateral postaxial polydactyl
Kocak <i>et al.</i> <sup>[9]</sup>	2015	4/male	Congenital	Not mentioned	Parietal and occipital/blond hair	None
		17/male	Congenital		Parietal and occipital/blond hair	None
		7/male	Since infancy		Right parietal/whitish color	None
		19/male	Since infancy		Vertex and left parietal/linear yellow circle All followed the Blaschko's lines	None
Dumitrascu <i>et al.</i> <sup>[5]</sup>	2016	4/female	Congenital	Not mentioned	Left-sided scalp/reddish color hair Right-sided scalp/brown color hair	Heterochromia of the eyelashes and Blaschkoid dyspigmentation of the skin
Douri <sup>[10]</sup>	2016	Case 1-10/male	Congenital	Not mentioned	Right part of the scalp/light brown hair	None
		Case 2-5/male			Right part of the scalp/light brown hair	None
		Case 3-9/male			Right part of the scalp/light brown hair	None
Kumar <sup>[11]</sup>	2017	6 months/male	Congenital	Omani origin	Midline in the occipital area/golden-yellow color	None

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### Conflicts of interest

There are no conflicts of interest.

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