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. Typically, colors such as brown, blond, red, and yellow hair presenting in the background of dark color hairs have been described in previous studies. 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, audiologic, 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...
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 eumelanic 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.\(^1\)\

There are various etiologies of scalp hair heterochromia, which includes genetic basis, metabolic defects, nutritional defects, or drugs [Table 1].\(^3\) 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.\(^1\)\(^,\)\(^2\)\

There are three different clinical types of asymmetrical scalp hair heterochromia: patchy, segmental, and diffuse type.\(^4\) 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).\(^3\)
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.\[2\] The review of literature of all the cases of patchy heterochromia of the scalp hair is given in Table 2.\[1-11\] 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.\[4\]

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 et al.\[6\]| 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 2, 3, and 5 followed the Blaschko’s lines                | None                                                                              |
| Iorizzo et al.\[7\]| 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                                             | None                                                                              |
|                    |      | Case 4-7/male    |                             |                | Occipital area/light brown color hair                         | None                                                                              |
|                    |      |                 |                             |                | All followed the Blaschko’s lines                             | None                                                                              |
| Qiao and Fang\[8\] | 2010 | 11/male         | Congenital                 | Chinese origin  | Vertex/brown color hair Consistent with Blaschko’s lines      | Lipedematous scalp                                                               |
| Zeng et al.\[9\]  | 2011 | 11/male         | Congenital                 | Chinese origin  | Vertex/brown color Showed arrangement in Blaschko’s lines on presented clinical picture |                                                                    |
| Bonamonte et al.\[10\]| 2014 | 5/male          | Congenital                 | Italian origin  | Numerous patches, especially over tempo-auricular and frontoparietal area | None                                                                              |
| Park et al.\[11\]  | 2015 | 8 months/male   | Congenital                 | Korean origin   | Left parietal scalp/brown                                     | Congenital bilateral postaxial polydactyl                                        |
|                    |      |                 |                             |                | Vertical band shaped following the lines of Blaschko           |                                                                    |
| Kocak et al.\[12\]| 2015 | 4/male          | Congenital                 | Not mentioned   | Parietal and occipital/blond hair                             | None                                                                              |
|                    |      | 7/male          | Congenital                 |                | Parietal and occipital/blond hair                             | None                                                                              |
|                    |      | 19/male         | Congenital                 |                | Right parietal/whitish color                                  | None                                                                              |
|                    |      |                 |                             |                | Vertex and left parietal/linear yellow circle                 | None                                                                              |
|                    |      |                 |                             |                | All followed the Blaschko’s lines                             |                                                                    |
| Dumitrascu et al.\[13\]| 2016 | 4/female        | Congenital                 | Not mentioned   | Left-sided scalp/reddish color hair                           | Heterochromia of the eyelashes and Blaschko dyspigmentation of the skin          |
| Dour\[14\]      | 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\[15\]     | 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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