

Isolated Patchy Heterochromia With Pili Annulati Features on Light and Electron Microscopy

Alejandro Lobato-Berezo¹, David Pesqué¹, Ramon M Pujol¹

¹ Department of Dermatology, Hospital del Mar-Parc de Salut Mar, Barcelona, Spain

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Corresponding author: Alejandro Lobato Berezo, M.D, Department of Dermatology 1.-Hospital del Mar, Barcelona, Passeig Marítim, 25-29, 08003 Barcelona, Spain. Telephone: (+34) 616717411 Fax: (+34) 932483328 E-mail: allobe@hotmail.es

Introduction

Isolated patchy heterochromia is a rarely described distribution of hair scalp mosaicism, where 2 distinct hair colors are present in the same person. We present the case of a patient with light microscopy and transmission electron microscopy (TEM) features that corresponded to pili annulati.

Case Presentation

A 12-year-old boy presented with a band of red hair over the interparietal region that was present since birth and had become more obvious over the years (Figure 1A). The rest of his scalp hair, eyebrows and eyelashes were brown. No previous traumas on the area, no treatments neither scalp inflammation were reported. The underlying scalp was normal. He had no diseases and the rest of physical exploration revealed no alterations. Light microscopy of both the red hair and brown hair showed light and dark segments under reflected light (Figure 1B). The latter corresponded to areas

of cavitation within the hair cortex which after exposure to a 10% potassium hydroxide solution, induced disappearance of the dark areas within 5 minutes (Figure 1C). In order to perform TEM, the hair samples were mounted on stubs and sputter coated with gold palladium. TEM of the hairs shafts showed a circular to oval cross-section. The cuticle consisted of a cobblestone pattern with overlapping cells. The microfibers composing the cortex of the red hair shafts presented large gaps between them (Figure 1D). The clinical findings could correspond to isolated patchy heterochromia with pili annulati microscopic features.

Conclusions

Heterochromia of the hair represents a variant of pigmentary mosaicism that can present in different distributions (patchy, segmental, diffuse or blaschkoid). First described in 2001 by Restano et al, isolated patchy heterochromia has been rarely reported, with less than 25 cases in the literature [1-3]. It is usually a benign condition not related to any

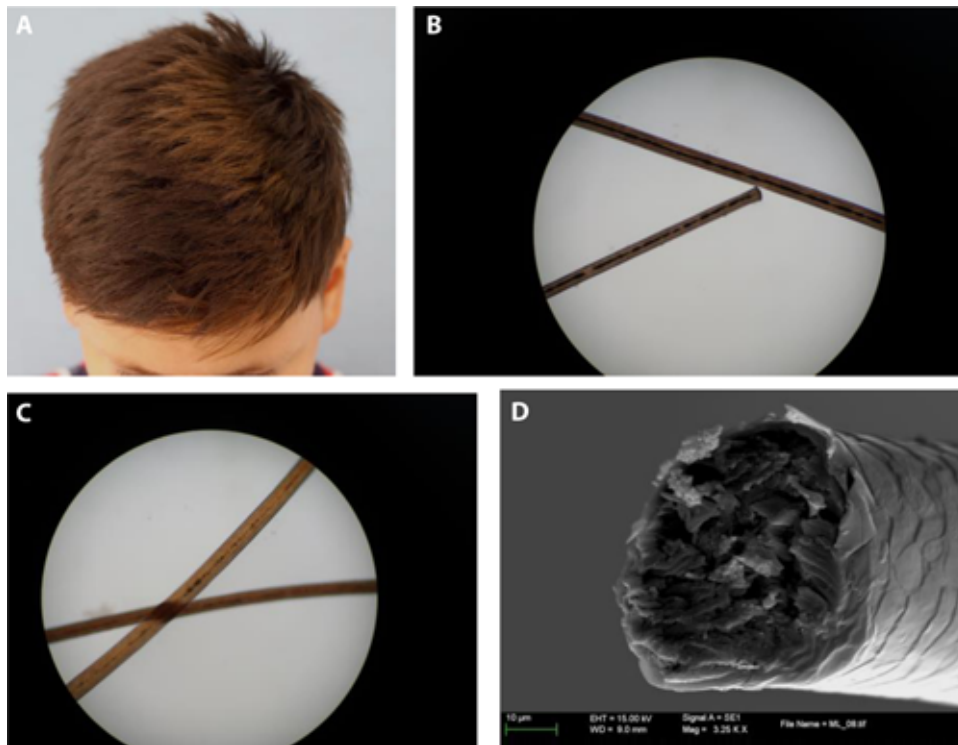


Figure 1. (A) Isolated patch of red hairs surrounded by a background of brown hairs. (B) Light and dark segments alternating in the hair shaft. (C) Progressive disappearance of the dark areas after 5 minutes of exposure to a 10% potassium hydroxide solution. (D) Multiples cavities in the cortex and cobblestone pattern in the cuticle.

genetic abnormalities of melanocyte migration (piebaldism, Waardenburg syndrome, Tietze syndrome, Vogt-Koyanagi-Harada syndrome), genetic diseases affecting pigmentation (tuberous sclerosis), inflammatory conditions damaging the melanocyte (vitiligo, halo nevus, regrowth in alopecia areata, herpes zoster, radiation), tumors (congenital melanocytic nevus), metabolic/nutritional defects (severe iron deficiency, vitamin B12 deficiency, kwashiorkor, phenylketonuria, Menkes syndrome), drugs (diazoxide, minoxidil, or chloroquine) or accidental causes (copper; green hair, cobalt; white-blue hair, tar; yellow hair, trinitrotoluene; reddish-brown hair) [2]. No structural abnormalities of the hair shaft are detected at light microscopy or transmission/scanning electron microscopy but for a smaller diameter of the lighter hairs when compared with the darker ones [1].

Pili annulati represents a hair abnormality of uncertain origin that is usually inherited in an autosomal dominant pattern [4]. It is characterized by alternating light and dark bands that can be seen with the naked eye, resulting in a speckled appearance to the hair [5]. As in our case, the light bands correspond to air-filled cavities located within the cortex of the hair shaft. Altered hair growth and fragility are not clinical features in this condition [5].

In conclusion, we report the first case of isolated patchy heterochromia with pili annulati microscopic changes on

light microscopy and TEM. These explorations should be routinely practiced to get a better characterization of hair heterochromia.

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