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Woolly hair nevus with associated nevomelanocytic nevus and attention-deficit hyperactivity disorder in a seven-year-old girl

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Abstract

Woolly hair nevus is a rare, non-genetically determined condition in which unruly and tightly curled hair is localized on one or several areas of the scalp. This may be associated with epidermal or melanocytic nevus. Patients presenting with this condition must be examined completely and additional ancillary procedures should be requested to determine associated anomalies and rule out the possibility of a syndrome. A regular follow-up is advised to determine if a case will progress into a syndrome.
We report a seven-year-old girl with woolly hair nevus with associated nevomelanocytic nevus and attention-deficit hyperactivity disorder. There were no associated ophthalmologic findings, cardiovascular abnormalities, and other systemic anomalies.

Keywords: Woolly Hair Nevus, Woolly hair, Woolly hair Nevus syndrome, attention-deficit hyperactivity disorder.

INTRODUCTION

Wooly hair nevus is a rare nonhereditary abnormality in the growth of hair which is characterized by tightly curled hair in one or more circumscribed areas of the scalp. This localized variant may be associated with extracutaneous anomalies. Patients presenting with this condition must be examined completely to rule out a syndrome associated with ocular anomalies, palmoplantar hyperkeratosis, heart abnormalities, retarded bone growth, tooth anomalies, and disturbance in the development of speech.

Case Summary

A seven-year-old girl born to non-consanguineous parents had a patch of curly hair with a nevomelanocytic nevus on the right parietal area of the scalp for three years. The patient had no growth retardation but was reported to have poor attention span in the school. There were no past illnesses that affected physical development. She was the only child and there was no similar hair condition noted up to the 4th genealogy.

Anthropometric examination revealed a height of 132.08 cm (2 score of 28.878) and a weight of 27 kg (2 score of 12.738) which were normal for her sex and age. Dermatological examination of the scalp showed a circumscribed 5 cm x S cm patch of extremely curly hair located over the right parietal region of her scalp. The hair shafts were nonfragile, kinky, and light brown in color. (Fig. 1). There was a solitary Smooth black flat-topped papule on the right parietal region of scalp. The examination of the rest of the scalp hairs was normal. The patient had no palmoplantar keratoderma or other skin lesions.

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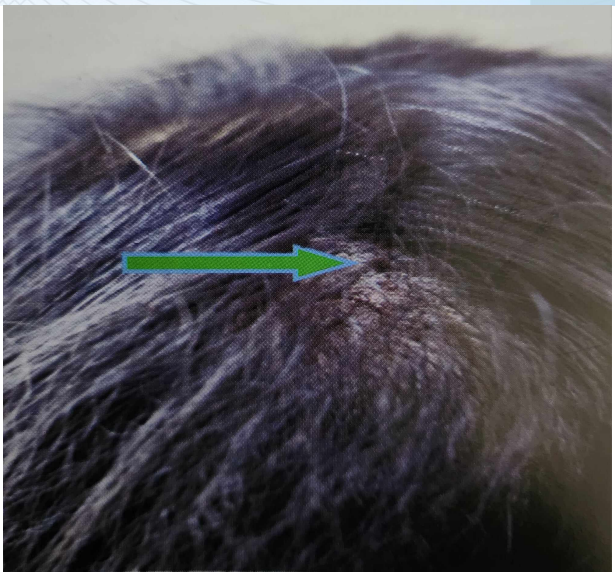

Figure 1. A circumscribed 5cm x 5cm patch of extremely curly hair located over the right parietal region of scalp. The hair shafts were nonfragile, kinky, and light brown in color. There was a nevomelanocytic nevus (green arrow) within the affected area.

Light microscopic examination of the hair revealed that the curly hairs were lighter and appeared smaller in diameter than the normal hairs (Fig.2)

Cardiac evaluation with 12-lead electrocardiogram revealed normal findings. Skeletal X-ray examination showed appropriate bone age without any osseous abnormality. Ophthalmologic evaluation showed normal ocular findings with visual acuity of 20/20. Psychomotor development evaluation by a pediatric neurologist revealed an attention-deficit hyperactivity disorder (ADHD), inattentive type.

Dental examination and evaluation was done to rule out delayed tooth eruption of the central top incisor. According to American Dental Association, the central top incisor is expected to shed at 6 to 7-years-old and permanent tooth is expected to erupt at 7 to 8-years-old. Given that the patient is only 7-years-old, the absence of her tooth is still within normal limits.

With all these findings, our patient was diagnosed clinically with woolly hair nevus with associated nevomelanocytic nevus and ADHD. There is no treatment for this condition at present. For the woolly hair nevus, we suggested the use of hair accessories such as hair clips, headband, and hair gels to hold, improve, and camouflage the hair appearance. The parents were counseled that although most cases of woolly hair nevus would run a benign course, monitoring for the development of possible extracutaneous anomalies should still be done. Behavioral therapy was given to address ADHD.

Discussion

Woolly hair was first described by Gossage in 1907 in a European family as a congenital structural anomaly of scalp hair that occurs sporadically or genetically. The term woolly hair nevus was first used by Dr. Fred Wise in 1927 when he reported two 5-year-old girls with a circumscribed area of light woolly hair on the scalp, while the rest of the hair was straight and brown. In comparison with the curled hair of dark-skinned individuals, the curls of woolly hair tend to merge rather than separate.10 Hutchinson et al have classified woolly hair types as follows: Type 1 which is autosomal dominant woolly hair (hereditary woolly hair), Type 2 which is autosomal recessive hereditary

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woolly hair (familial woolly hair), Type 3 which is symmetrical circumscribed allotrichia, and Type 4 which is woolly hair nevus. Post, on the other hand, suggested a classification of woolly hair nevus into Type 1 which has no associated disorder in the glabrous skin or scalp; Type 2 which has associated linear verrucous nevus; and Type 3, also known as acquired progressive kinking of scalp hair in young adults with short, dark, kinky hair.

Our patient was classified as woolly hair nevus Hutchinson type 4/ Post type I. Unlike the inherited variants (Hutchinson types 1 and 2) which are considered generalized forms and clinically presents with involvement of the entire scalp, 2022 our patient only had a limited circumscribed area affected, unlike in Hutchinson type 3 wherein the woolly hair is symmetrically circumscribed.

Woolly hair nevus is characterized by one or more well- circumscribed areas of the scalp of curly, kinky, shiny hair, of woolly texture, with onset within the first two years of life. Our patient's hair abnormality was noted at the age of four. The hair usually grows normally and is difficult to comb but not fragile. l The size of affected area generally grow in proportion with the body, and may reach a certain size then remain constant throughout life. There is no sexual preponderance.

Histologic findings are varied in the literature. Ll Light microscopy reveals the affected hair to be thinner, more flat and less pigmented than normal hair. il Woolly hair nevus accompanied by ipsilateral linear epidermal nevus is common and seen in nearly 50% of the cases. 1415 In our case, there was a nevomelanocytic nevus within the affected area. Extracutaneous abnormalities have been described, especially when the woolly hair nevus is associated with an epidermal nevus, constituting an epidermal nevus syndrome. In those cases, it is important to rule out further compromise of the ophthalmologic, nervous, and cardiac systems, 411.17

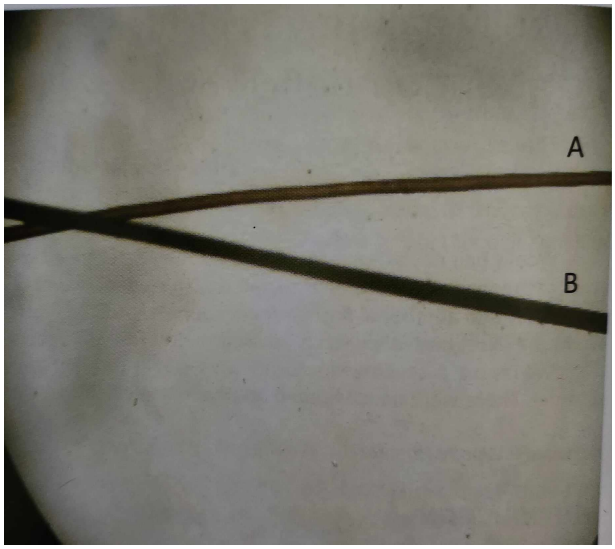

Figure 2. Light microscopic examination of the wooly hair (A) and normal hair (B).

Woolly hair nevus has been reported to be associated with retinal anomalies, retarded bone growth, teeth abnormalities, and disturbances in speech development.* In 1988, the first association with cardiac abnormality wa detected in an electrocardiographic study of two affected children. After four years of follow-up, the patients developec dilated echocardiographic cardiomyopathy and later died c cardiac failure.

In our case, thorough physical examination and systemic evaluation revealed normal findings except for the incidenta finding of ADHD. This association has not been previously reported in the literature.

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Conclusion

Our patient had woolly hair nevus with associated nevomelanocytic nevus and attention-deficit hyperactivity disorder. Though most cases run a benign course, the patient should still be monitored to determine occurrence of other associated anomalies and development into a syndrome later

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