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CASE REPORT

Exuberant generalized hypertrichosis in an infant: case report

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Abstract

Hypertrichosis is characterized by excessive hair growth in any body region, occurring either in a localized or generalized manner. The last one is commonly associated with genetic syndromes or use of specific drugs. This case report presents an infant with mucopolysaccharidosis type II who underwent a bone marrow transplant (BMT) and received cyclosporine, consequently exhibiting an exacerbated condition of generalized hypertrichosis. The aim of this study is to review the main causes of hypertrichosis and discuss hypotheses regarding the development of this pathology.

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CASE REPORT

A 1-year-and-5-month-old male infant was taken to the dermatology clinic for mild atopic dermatitis on his face. He had his IDS gene sequenced at birth and was diagnosed with mucopolysaccharidosis type II. Six months before, he had had a bone marrow transplant. He was taking carvedilol 2.5 mg/day, cyclosporine 8 mg/kg/day, magnesium 200 mg/day, trimethoprim-sulfamethoxazole 200 mg/day, acyclovir 400 mg/day, folic acid, a multivitamin, and prednisolone 0.1 mg/kg/day as part of a weaning protocol. He had no other comorbidities, and his gestational history was uneventful.

A physical examination revealed eczematous plaques on the face and dry skin across the body, along with generalized hypertrichosis, which was more pronounced in the lateral regions of the lower limbs, the extensor surfaces of the upper limbs, the back, and the face (Figures 1 and 2). The mother noted that the patient had experienced hypertrichosis since birth, but it was not as prominent and worsened following the bone marrow transplant. She provided laboratory tests conducted the day before, which showed TSH levels of 2.16 µUI/mL and free T4 levels of 0.87 ng/dL, both normal for his age, ruling out the possibility of hypothyroidism.



Figure 1. Hypertrichosis in the back and extensor surface of the arms.



Figure 2. Hypertrichosis on the forehead and glabella. Coarse facial features, a depressed nasal bridge, a broad base of the nose, prominent ears, and a short neck.

DISCUSSION

Hypertrichosis is characterized by excessive hair growth anywhere on the body, unlike hirsutism, which refers to increased hair growth in areas of the skin influenced by androgenic hormones[1,2]. It can be categorized as generalized or localized[1]. The generalized form may result from various genetic syndromes[1], such as mucopolysaccharidosis type II[3], and it can also occur as a secondary effect of certain medications[1,2]. Some drugs most commonly linked to generalized hypertrichosis include valproic acid, phenytoin, penicillamine, cyclosporine, and maternal use of minoxidil[1,4]. Our patient exhibited both the genetic syndrome and exposure to cyclosporine, which is known to cause hypertrichosis. We suspect that the primary factor contributing to hypertrichosis is exposure to cyclosporine since the condition was significantly milder before medication was started.

Cyclosporine is a lipophilic cyclic polypeptide that inhibits the transcription of interleukin-2 in helper T lymphocytes [2,5]. This action prevents the activation and proliferation of helper T lymphocytes (CD4) and cytotoxic T cells (CD8), thereby inhibiting critical steps in the cellular immunity pathway[5].

Consequently, cyclosporine possesses immunosuppressive properties and is utilized as first-line therapy for the prophylaxis and treatment of transplant rejection[5], particularly in bone marrow transplants.

Hypertrichosis is one of the most common adverse effects associated with cyclosporine use, but the symptoms are generally mild to moderate and resolve with a reduction in the drug dose[5]. Approximately 50% of patients experience reversible hypertrichosis, particularly on the face and upper trunk[4]. Its pathogenesis is not fully understood, but it is known to be a dose-dependent side effect observed in the pediatric population, mainly within the first six months of treatment[2]. Our patient was in his sixth month of cyclosporine therapy, consistent with the literature. However, no improvement in hypertrichosis was noted with the gradual reduction of the drug dose, suggesting that the persistence of symptoms may be related to both the drug use and the presence of mucopolysaccharidosis type II.

Additionally, when high doses of oral glucocorticoids are administered over extended periods, androgen-independent hair grows, primarily on the backs of children[4]. This is likely due to the direct conversion of hair from the telogen phase to the anagen phase[4]. The patient has been gradually reducing his prednisolone dosage since the bone marrow transplant, which also raises the possibility that the medication may influence the onset of hypertrichosis.

The case presented illustrates an exuberant depiction of generalized hypertrichosis, suggesting that multiple factors may have contributed to its occurrence. It is essential to understand the most common causes of hypertrichosis in children and recognize that there may be additional effects from various causal agents in some instances.

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