Hypertrichosis Induced by Diazoxide in Idiopathic Hypoglycemia of Infancy

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Case Report

C... Laura, the product of an uneventful pregnancy, was born on February 8, 1988. She weighed 4.2 kg and looked healthy. However, during the first days of life, she developed severe hypoglycemia, responsive only to continuous intravenous glucose and glucagon infusion. High plasma insulin, inappropriate for the low blood glucose levels, was found.

On day 20, diazoxide treatment was started (20 mg/kg/day p.o.). Five weeks later, hypertrichosis developed on the forehead, cheeks, back and extensor aspect of the limbs. There was not only vellus hair hypertrophy or hypertrichosis lanugosa but also true hair, 2 cm long, rather thick, brown-black (fig. 1). The hyperinsulinemia persisted in spite of the therapy, and it proved impossible to withdraw glucagon; diazoxide was stopped in mid-april and an 85% pancreatectomy was performed on August 24, 1988. The histology of the pancreas showed the presence of several hyperplastic foci of B cells. After withdrawal of diazoxide, hypertrichosis faded slowly; 6 months later, the skin looked almost normal.

Before, during and after the treatment, the secondary sexual characters remained quite normal; hirsutism, external genitalia hyperplasia and breast development were not observed at any time; urinary 17-keto- and 17-hydroxysteroids fell within the range of normal values.

Comment

Diffuse hypertrichosis is now well recognized as a side effect of diazoxide treatment [1, 2]; it occurs more frequently in idiopathic hypoglycemia than in hypertension where the overall incidence is lower than 1% [1,2].

Hypertrichosis develops very rapidly, a few weeks only after initiation of the treatment, and becomes more and more impressive with time. Fortunately, when the drug is withdrawn, hypertrichosis fades progressively with restitutio ad integrum within 6 months. Histology reveals a high proportion of anagen hair follicles [3, 4].

Clear-cut etiopathogeny escapes present knowledge, but it has been hypothesized that vasodilatation increases cutaneous perfusion and promotes hair growth; a rich vascular network around hair follicles was recognized under the microscope [1,2,4];

inhibition of phosphodiesterase induces an increase of cyclic AMP within the hair bulb and promotes cell proliferation [4]; even if circulating gonadal and adrenal androgens stay within physiological limits, a specific property of the drug may induce the cutaneous 5α -reductase and thus increase the androgen production of hair bulbs [5, 6].

Anamnestic data allow to differentiate hypertrichosis induced by diazoxide from other iatrogenic conditions (e.g. minoxidil, diphenyl hydantoin) and from congenital and hereditary disorders (e.g. Cornelia de Lange syndrome).

Fig. 1. Hair growth was especially conspicuous on the forehead, temples and cheeks.

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