Case Report

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Hairy Pinnae after Orchiectomy and Chemotherapy for Testicular Cancer: Acquired Localized Hypertrichosis of the Ears

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Key Words
Cancer therapy · Chemotherapy · Hypertrichosis · Orchiectomy · Pinnae

Abstract
Acquired localized hypertrichosis has rarely been reported. Here, we describe a patient with localized hypertrichosis of the pinnae that occurred 4 months after orchiectomy and chemotherapy for a testicular carcinoma. To our knowledge, this is the first case of an acquired hypertrichosis of the pinnae after cancer therapy. We propose that in our patient either hypogonadism or the hormonal imbalance caused by the cancer therapy led to the development of the hairy pinnae, perhaps alongside a genetic predisposition for hairy ears.

Introduction

Acquired localized hypertrichosis has rarely been reported. Here, we describe a patient with localized hypertrichosis of the pinnae that occurred after orchiectomy and chemotherapy for a testicular carcinoma.

Case Report

A 46-year-old man was referred for evaluation of a striking pilosity on his ears, which developed after therapy of a mixed germ cell tumor of the right testicle. The patient underwent surgery and chemotherapy consisting of 4 cycles of etoposide, cisplatin and bleomycin in combination with courses of filgrastim (GCS-F). There was no family history of anomalous hair growth. The patient experienced complete hair loss after the first cycle of this regimen. Four months after chemotherapy, vellus hair started to grow again, and hair growth also extended to his ears, and transiently to his back and shoulders. Six months after therapy, his hair returned to its former density, consistency and color, but the excessive pilosity on his ears transformed into terminal hair and persisted. One year after chemotherapy, the patient was given subcutaneous testosterone injections due to symptomatic reduced testosterone levels. The testosterone dose is currently 1,000 mg once every 6 weeks, without any obvious impact on hair growth.

On examination, terminal hair growth was particularly prominent on the middle and lower ear helix and anthelix, with the scapha, tragus, antitragus and lobule also affected (fig. 1, 2). Vellus hair growth was sparse. The rest of the physical examination revealed a discrete diffuse palmoplantar keratosis. This had been present since adolescence, and the patient had previously received acitretin for it. The patient was otherwise in a generally good condition, and without any evidence of a relapse of his disease. Hormone status 2 years after chemotherapy was as follows: free testosterone 38.7 pmol/l (reference interval, RI, 25–80 pmol/l), testosterone 13.8 nmol/l (RI 8.4–28.7 nmol/l), estradiol 0.11 nmol/l (RI 0.07–0.18 nmol/l), prolactin 405 mU/l (RI 75–400 mU/l). FSH concentration was increased at 32.2 IU/l (RI 1.6–18.1 IU/l), and the LH level was 23.9 IU/l (RI 2.0–18.0 IU/l). Recent laboratory findings were normal, except for a deficiency in 1,25-dihydroxy-vitamin D (38 nmol/l, RI 75–220 nmol/l) and an elevated level of hemoglobin at 182 g/l (RI 135–175 g/l). As treatment for the localized hyperpilosity we proposed laser epilation.

The patient presented with an acquired localized hypertrichosis that only permanently affected the pinnae. Physiological growth of coarse terminal hair on the pinnae is found in males at variable frequencies in distinct populations, and with a variable age of onset. It is probably a Y-linked inheritable trait, although recent

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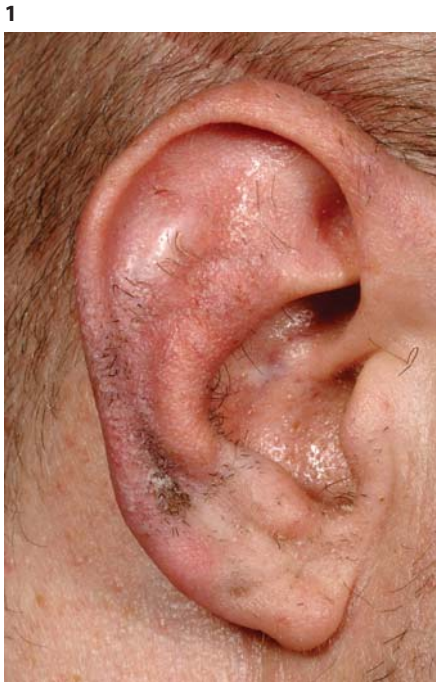

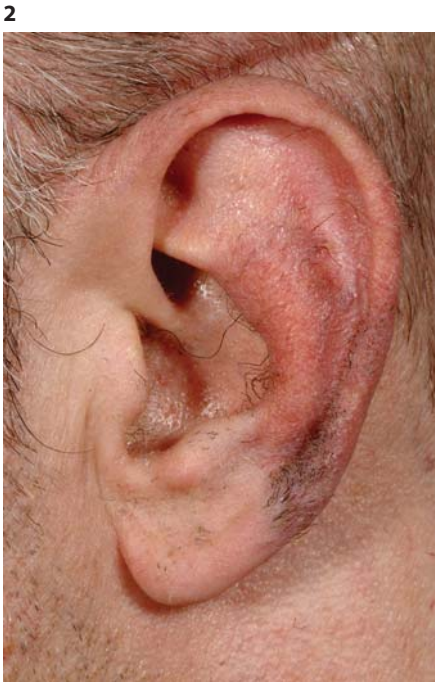

Fig. 1. Right ear of patient with hypertrichosis (2010).
Fig. 2. Left ear of patient with hypertrichosis (2010).

reports question this contention [1–3]. In our case, the symmetrical pattern of hair growth, with hypertrichosis of the middle and lower ear helices, resembles this trait. However, before cancer therapy, the pinnae showed no terminal hair growth and the family history was negative.

Discussion

There are few reports about pathological localized hypertrichosis of the ears. Sometimes it may be related to an underlying malignancy. In our patient, there was no evidence of a relapse of his testicular tumor at time of presentation. Furthermore, the clinical features were not consistent with paraneoplastic hypertrichosis lanuginosa. In the latter, growth of mostly vellus hair characteristically appears before the discovery of the tumor and may regress after tumor removal [4]. Hairy pinnae have been described in babies with XYY syndrome and babies of diabetic mothers [5]. Furthermore, Tosti et al. [6] described a patient with AIDS who developed both trichomegaly of the eyelashes and hypertrichosis of the ears. Our patient was HIV-negative, and had no signs indicating an underlying immunodeficiency or metabolic disease.

Hypertrichosis has been associated with the intake of a number of drugs, such as phenytoin, acetazolamide, streptomycin, latanoprost, cyclosporine, psoralen, diazoxide and minoxidil [7]. However, in these cases, the acquired changes in hair density, texture and structure usually affect large body areas and disappear upon discontinuation of the drug. Treatment with IFN-α can induce trichomegaly and hypertrichosis of the eyebrows [8]. Finally, hypertrichosis after chemotherapy is a rare phenomenon. George and Whitton [9] described a woman with breast carcinoma who received radiotherapy and chemotherapy with Adriamycin and cyclophosphamide. After treatment, her scalp hair regrew, but hairs also appeared on her neck and cheeks. In one case, baldness was reversed shortly after chemotherapy with cisplatin and docetaxel [10]. Using Medline, we were not able to find localized hypertrichosis associated with either bleomycin, etoposide, cisplatin or filgrastim treatment.

Hair modification has recently been described in up to 90% of the patients treated by epidermal growth factor receptor inhibitors. In addition to alopecia and texture/color changes, patients complain of slow scalp hair growth, facial hair growth [11] and trichomegaly [12]. In one case, gefitinib therapy resulted in striking terminal hair growth on the nose tip [13], whereas in a formerly bald patient hypertrichosis of the scalp, eyelashes and eyebrows occurred after cetuximab therapy [14]. It is interesting that in body areas affected by pathological localized hypertrichosis – such as the ear and nose, and to a lesser extent, face and neck – mantle hairs occur in large numbers. The function of these pilosebaceous follicles is unknown [15]. It can be inferred from the morphology of a persistently dormant state, both of the follicular epithelium and sebaceous gland, that they serve a specific function, e.g. as a protective hood for nerve endings. In this way, they physiologically evade the common hormonal regulation of hair follicles. Disruption of an inhibitory regulative element – overriding growth stimulation or regulation that is specific to them – might play a common role in the pathogenesis of hypertrichosis of the pinna in our patient and the cases cited above.

So, there are two possible mechanisms of hair growth stimulation that may have led to the hairy pinnae. First, a direct action of the chemotherapeutical agents used in our patient. However, the observation that there was a gradual coarsening of the vellus into terminal hair long after chemotherapy stopped, while newly grown body hair shed again, counts against this

possibility. Second, considering the hypogonadism – confirmed 1 year after cancer therapy but obviously present immediately after treatment – one can speculate that low testosterone levels combined with a compensatory increase in GnRH and LH/FSH may have played a role. It is known that the root shaft of anagen hair expresses LH and hCG receptors [16]. At least in animals, GnRH and gonadotropins have been implicated in increased hair growth after spaying [17].

It is thus tempting to speculate that in our patient either hypogonadism or the hormonal imbalance caused by the cancer therapy accelerated the development of hairy pinnae; this may have occurred alongside a potential genetic predisposition for hairy ears. To our knowledge, this is the first case of an acquired hypertrichosis of the pinnae after cancer therapy. Further investigations are required to establish whether hypertrichosis of the ears in older men is linked to a change in sex hormones, and to assess the role of mantle hairs in the development of acquired localized hypertrichosis.

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