



Anterior cervical hypertrichosis: a sporadic case

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Abstract

Anterior cervical hypertrichosis is a very rare form of primary localized hypertrichosis. It consists of a tuft of terminal hair on the anterior neck just above the laryngeal prominence. The etiology is still unknown. In this article, we reported a 15-year-old female patient who presented to our clinic with a complaint of hypertrichosis on the anterior aspect of the neck for the last five years. Her past medical history revealed no pathology except for vesicoureteral reflux. On the basis of clinical presentation, our patient was diagnosed with anterior cervical hypertrichosis and she was considered to be a sporadic case due to lack of other similar cases in familial history. To date, 33 patients with anterior cervical hypertrichosis have been reported. Anterior cervical hypertrichosis can be associated with other abnormalities, but it frequently presents as an isolated defect (70%). The association of vesicoureteral reflux and anterior cervical hypertrichosis which was observed in our patient might be coincidental. So far, no case of anterior cervical hypertrichosis associated with vesicoureteral reflux has been reported in the literature.

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Keywords: Anterior cervical hypertrichosis, sporadic, vesicoureteral reflux

Introduction

Anterior cervical hypertrichosis (ACH) is a very rare form of primary regional hypertrichosis characterized with a tuft of terminal hair on the anterior neck just above the laryngeal prominence (1). The etiology is unknown (2).

Case

A 15-year old girl presented to our clinic with a complaint of hypertrichosis on the anterior aspect of the neck which had been lasting for the last five years. The patient was born by normal vaginal delivery at term. Her past medical history revealed no pathology except for vesicoureteral reflux (VUR). There was no consanguinity between her mother and father. Voiding cystoureterography revealed grade 1 VUR on the left side and dimercaptosuccinic acid (DMSA) renal scintigraphy revealed a reduction in the total activity in the left kidney (cortical hypofunction) (Figure 1). The patient's height was measured to be 150 cm and her weight was measured to be 43 kg. Her familial history revealed no pathology. On physical examination, multiple terminal hair were observed on the anterior aspect of the neck just above the laryngeal prominence (Figure 2, 3). There was no hypertrichosis in the other body regions except for axillary and pubic hair which are expected findings

in the adolescence. Among the laboratory variables ordered for investigation of hyperandrogenemia, DHEA-SO₄, free-total testosterone and surrenal gland ultrasonography were found to be normal. On the basis of the clinical findings, a diagnosis of ACH was made and the patient was considered to be a sporadic case due to lack of other similar cases in familial history. Ophthalmologic and neurological examination performed to investigate accompanying anomalies were found to be normal. Brain magnetic resonance imaging and electroneuromyography were found to be normal. Plain and lateral cervical graphies revealed no pathology except for a longer than normal C7 transverse prominence on the right side. The patient did not accept biopsy. Laser epilation was offered and a marked reduction was observed in the hair after three applications (Figure 4). Written informed consent was obtained from the patient's family.

Discussion

Anterior cervical hypertrichosis was reported in three patients by Trattner et al. (3) for the first time. These patients were members of Arabic families (two generations) with consanguineous marriage. Peripheral neuropathy and bilateral hallux valgus were present in all three patients. Bilateral optic nerve atrophy and macular dysfunction were found in one patient. Tsuka-

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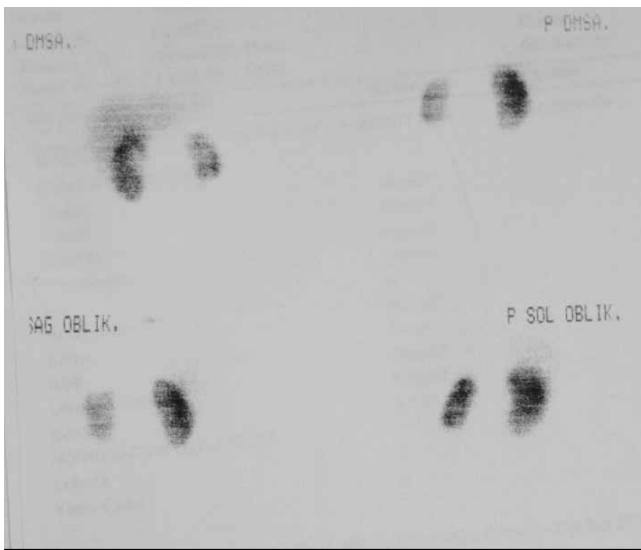


Figure 1. Reduction in total activity in the left kidney on DMSA renal scintigraphy (cortical hypofunction)
DMSA: Dimercaptosuccinic acid



Figure 3. Lateral view of the patient



Figure 2. Multiple terminal hair on the laryngeal prominence



Figure 4. Reduction (70%) in the hair after laser epilation

hara and Kajii (4) reported ACH in seven members of a Japanese family (three generations). It was reported that no other pathology accompanied in these patients except for one patient who had Turner's syndrome. It has been reported that anterior cervical hypertrichosis may be observed in an age range varying from birth to early childhood (5). Familial and sporadic cases have been reported. The inheritance patterns are different in hereditary cases. It generally has an autosomal dominant inheritance, but autosomal recessive and X-linked

dominant inheritance have also been reported (4, 6). There are also sporadic cases without a positive familial history in the literature (2). Our patient was considered to be a sporadic case due to lack of a positive familial history. Up to the present time, 33 cases of anterior cervical hypertrichosis (alone or accompanying different anomalies) have been reported. It frequently occurs as a solitary defect (70%). The most common accompanying anomaly is peripheral sensory and motor neuropathy (70%). The other anomalies which may accompany in-

clude hallux valgus (50%), ophthalmological anomalies (optic atrophy, chorioretinal changes) (40%), mental retardation (30%) and hypertrichosis on the back (20%) (5). Hypertrichosis is a cosmetic problem which leads to stress. Permanent treatment options include electrolysis, laser epilation and Intense Pulsed Light treatment, but the method which is recommended most commonly is laser epilation (7). Laser epilation was applied to our patient and it was found that a 70% reduction occurred in the hair.

In this article, a case of ACH accompanied by VUR has been presented. Up to the present time, no case of ACH accompanied by VUR has been reported in the literature. The association of VUR and ACH found in this patient may be coincidental. However, radiological, neurological and ophthalmologic examinations should be performed in all patients in addition to detailed physical examination, because ACH may accompany many anomalies as stated in the literature.

Informed Consent: Written informed consent was obtained from patient's parents who participated in this study.

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