A novel finding of hair growth like vellus hairs on glabrous skin of distal phalanx of thumb in Vogt-Koynagi-Harada Disease

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Title:

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Abstract:

This article reports a case of a 25 year old female patient who came with an unusual complaint of growth of hair on palmer surface of thumb and on side of nail after two year of Vogt-Koynagai-Harada disease (VKHD). It was published because of unusual presentation mimicking optic neuritis first and later developing into bilateral pan uveitis with serous retinal detachments and alopecia. The hairs were fine and short which lasts for 4-6 weeks. The presence of hairs at this unusual site is a novel finding. The same patient came again with the complaint of hair growth on palmer surface of distal phalanx of thumb after two years which lasts for 4 to 6 weeks.

Key Words: Vogt-Koynagi-Harada Disease, Vellus hair, Alopecia, Tinnitus

Key clinical message:

Vogt-Koyanagi-Harada Disease (VKHD) is a systemic autoimmune condition that affects melanocyte-rich tissues, leading to ocular, auditory, and skin-related complications. While hair loss, vitiligo, and poliosis are recognized features of VKHD, the emergence of vellus hair on the normally hairless skin of the distal thumb is an unusual and previously undocumented occurrence. This case emphasizes the possibility of immune system dysregulation in VKHD contributing to hair follicle activation in unexpected areas, broadening our knowledge of its dermatological effects.

Introduction:

Alfred Vogt in 1906 followed by Koynagi in 1911 and then Harada in 1923 published cases with description of VKHD. Harada described the posterior disease as acute posterior choroiditis.[1] It is a multisystem autoimmune disorder that affects melanocyte-containing organs such as the eye, central nervous system, skin and auditory system.[2] Age group is between 20 to 50 years, but it may occur in children and in old age.[3,4]

In the acute VKHD there are prodromal symptoms such as headache, tinnitus or vertigo followed by non-granulomatous inflammation involving the whole uveal tract causing sudden visual deterioration. Bilateral multiple serous retinal detachment (SRD) with variable severity developed.[5]

VKHD is associated with various skin findings in convalescent stage such as vitiligo of eyebrows, eyelashes and skin. Alopecia of scalp hair may occur temporarily.[6]

Israelsen NM et al, a dermatologist by using ultrahigh resolution optical coherence tomography (OCT) detected vellus hairs on glabrous area of palm which were comparable with vellus hairs on cheek.[7] This study provides the basis to report the finding which we observed in our patient.

Case Presentation:

This is a case that has been reported previously in 2017 by me and published in CPSP journal.[5] A 25 year old female presented with the history of prodromal symptoms of headache and neck stiffness. On examination there was bilateral optic disc hyperemia and swelling mimicking the picture of optic neuritis. The patient later developed bilateral pan uveitis with SRD. The case was treated successfully with posterior subtenon injection (PSTI) of triamcinolone. Later on in this patient an unusual finding developed which I am going to report for the first time. She came with the complaint of hair growth on radial and palmer surface of distal phalanx of thumb after two years in October 2019. Some of the hairs were near mucocutaneous junction. These hairs were fine, thin like vellus hairs. (Fig. No.1) Dermatologist was consulted for these integumentary changes. There was no history of hirsutism. This unique finding of vellus hairs came as a surprise as on extensive search of literature no such finding has been reported as yet in VKHD. She also developed tinnitus along with development of vellus hairs. The patient was on low dose steroids, prednisolone 5mg /day. The auditory symptoms were relieved by increasing the dose to 10mg per day. Whenever we stopped steroids the auditory symptoms recur and so the patient was kept on 5mg prednisolone per day for further two years. She didn't develop glaucoma or cataract during treatment.

She again developed hair in June 2021 and came twice to show these hairs (Fig.No. 2). These were very fine short hair. The hair remained for 4-6 weeks. After that she never complained of hair growth again.

Differential diagnosis :

Physicians must consider differential diagnosis of Vogt-Koynagi-Harada Disease in patients presenting with atypical signs and symptoms to make an early and accurate diagnosis before treating the ailment carefully. Sacrcoidos and alopecia areata are some immune mediated disease that must be considered as differential diagnosis. Endocrine and metabolic causes like hypothyroidism and Cushing's syndrome, along with medication-induced or post-inflammatory hypertrichosis, should also be considered. Additionally, genetic conditions like piebaldism and neoplastic syndromes such as Langerhans cell histiocytosis may present with similar findings.

Discussion:

Glabrous skin of palm and the papillary ridges is characterized by absence of hair follicles and sebaceous glands. Hairy skin complications are frequent in VKHD. Integumentary changes like alopecia, poliosis and vitiligo are late complications in this disease. The presence of vellus hairs on palm similar to cheeks have been demonstrated.[7] Melanocytes are found in meninges and inner ear in addition to eye, skin and hairs. The pathogenesis of VKHD though not completely understood, involves auto antigens, the melanocyte differentiation proteins, like tyrosinase (TYR), tyrosinase-related peptide (TRP)-1, TRP-2.[8,9] These proteins are the enzymes involved in the synthesis of melanin and are present specifically in melanocytes.[10] Additional auto antigens, like KU-MEL-1 and lens epithelium-derived growth factor have a role through an IgG-mediated mechanism.[9] The genetic predisposition involves certain genetic or epigenetic factors resulting in immune dysregulations.[11] The growth of hairs on palmer surface of distal phalanx of the thumb may be the result of auto antigens along with immune dysregulations.

Methodology, Project administration, Resources, Supervision, Validation, Visualization, Writing - original draft

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Data Availability Statement :

The data can be requested upon reasonable request from the corresponding author to maintain privacy.

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Consent Statement :

Written informed consent was taken from the patient.

Ethics Approval :

This does not apply to this study since this is a case report however our institute's quality service has authorized publication of this case.

Conflicts of interest :

There are no conflicts of interest to declare.

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