CASE REPORT

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Two Cases with Aquagenic Palmoplantar Keratoderma

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ABSTRACT Aquagenic Palmoplantar Keratoderma (APK) is a rare case characterized by edematous, white and wrinkling plaques on the palm after a short-term contact with water. Two male cases, aged 29 and 21, referred to our polyclinic at different times with a complaint of whitening which developed on the hands after a short contact with water. The cases were clinically diagnosed with APK and in one of the cases the diagnosis was supported with biopsy. Since both of the cases were asymptomatic, they were followed up with no treatment. Two male cases who had this rare disease which affects women more frequently were presented and APK literature was reviewed.

Keywords: Keratoderma; palmoplantar; male

quagenic Palmoplantar Keratoderma (APK) is a rare palmoplantar keratoderma which occurs after short-term contact with water. It was first defined by English et al. in 1996 with the name "transient reactive papulotranslucent acrokeratoderma". Later, it has also been named as aquagenic palmoplantar keratoderma, aquagenic syringeal acrokeratoderma, acquired aquagenic keratoderma, hereditary papulotranslucent acrokeratoderma. ²⁻⁴

In this paper, two cases aged 29 and 21 years, with the diagnosis of APK, are presented and the etiopathogenesis, diagnosis and the treatment of the disease are reviewed with literature. The consent form was taken from the patients.

CASE REPORTS

CASE 1

29-year-old male patient referred with a complaint of whitening that occurred after immersing his hands in water, which he had been aware for 10 days. The patient who stated that the complaint disappeared completely 25-30 minutes after the hands were taken off the water did not have any symptoms such as pruritus, burning or sweating and his feet did not show a similar complaint. Our case did not have any other diseases or a history of using drugs. Dermatologic examination showed mildly swollen white plaques which were intense in the palmar region of the hand, especially on the fingers, after the hands were immersed in water for two minutes (Figure 1a). The same findings were also seen on the soles of the feet after the feet were immersed in water (Figure 1b). Systemic examination did not reveal

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any findings of cystic fibrosis. Laboratory examinations including biochemical analysis of serum, hemogram and thyroid tests were within normal limits. Serologic test for syphilis was found to be negative. Punch biopsy sample was taken from the hand in the active period. Biopsy results showed compact ortokeratosis on the surface, mild dilatation in eccrine glands and capillarity increase around eccrine glands in the dermis (Figure 1c) These findings were consistent with aquagenic palmoplantar keratoderma. The patient was diagnosed as aquagenic palmoplantar keratoderma clinically by the help of histopathological findings.

CASE 2

21-year-old patient referred to our out-patient clinic with the complaint of white spots on his hands after being immersed in water which started a week ago. As in the other patient, this case also did not have complaints of pruritus, burning, sweating and family history. The dermatological examination of the patient whose anamnesis did not show any diseases or drug use showed intense white plaques on the palmar region of the hands, especially on the tips of fingers after the hands were soaked in water for two minutes (Figure 2). The lesions di-

sappeared completely about 20 minutes after the hands were dried. Laboratory examinations including complete blood, biochemical analysis of serum and thyroid tests were normal and syphilis serology was found to be negative. Since the case was asymptomatic, the patient was followed with no treatment.

DISCUSSION

APK is a rare, acquired clinical figure which affects adolescents and young adult women mostly. 1,5 APK is generally characterized by hypopigmentation, evident sulcus, development of numerous symmetric, smooth-round, transparentwhite papules and plaques on the palmar region and rarely in the plantar region, a few minutes after contact with water. Burning, pruritus, pain and hyperhidrosis can also accompany the clinical figure. The most important characteristic of the disease is the spontaneous recovery of the lesions a short while after the skin gets dry. 4,6 Neither of our cases had burning, pruritus, pain or hyperhidrosis. Palmoplantar involvement was found in Case 1, while only palmar involvement was found in Case 2.

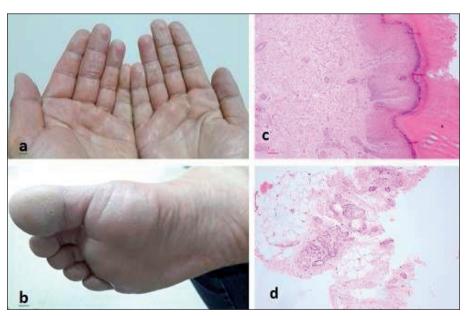


FIGURE 1: a) Milldy swollen white plaques which were intense in the palmar region of the hand, especially in the fingers, b) white plaques on the sole, c) compact ortokeratosis on the surface, mild dilatation in eccrine glands, d) capillarity increase around eccrine glands.



FIGURE 2: White plaques in the plamar region of the hand, especially in the fingers.

Although APK is a disease of young adult and adolescent women, there are also a few male cases in literature. It can show autosomal dominant and autosomal recessive inheritance, acquired cases have also been reported.³ Both of our cases were young and male. Yan et al. defined the appearance of lesions by keeping the hands in water as hand in the bucket sign and stated that it was an important sign in the diagnosis of aquagenic palmoplantar keratoderma². Hand in the bucket sign is the thickening and wrinkling of the skin and the formation of superficial, white papules on the palmar region, interdigital areas and lateral and upper sides of the hand after keeping in water for about two minutes.

Although its etiopathogenesis is not clear, it has been suggested that it can develop due to the structural and functional defect of the corneum layer during the adolescence and that it can be a primary disease of the sweat gland ductus.^{7,8} Other reported mechanisms are increase in water retention capacity in corneum layer as a result of the increase in sodium concentration, impairment in the barrier function of corneum layer as a result of keratin defect and increase in water absorption. In addition, developing reactions to some drugs is another emphasized mechanism.^{5,7,8} It has also been reported that there may be an association between cystic fibrosis, asthma, allergic rhinitis, palmar erythema, malign melanoma and COX-2 inhibitor use and APK.5,7 Systemic examination of our patients did not show any signs of disease or any drug use.

Histopathology is frequently nonspecific; however, may be performed to support the diagnosis, as we did in our first case. Histopathological changes are reported as follow; increased thickening of stratum corneum and ortokeratotic hyperkeratosis with abnormal staining, acrosyringium dilatation and hyperplasia at dermal eccrine glands, changes at eccrine cuboidal cells and increase in capillarity around and next to eccrine glands with vacuolization, many of which were seen in our Case 1.9

In the treatment of APK, 20% aluminum chloride solution, botulinum toxin injection, antihistamines, pomads including 5% salicylate and creams including 20% urea are frequently considered. Among these, aluminum salts are widely preferred treatment of choice. Akpolat et al. applied 20% aluminum chloride to six cases and at the end of two weeks, complete response was obtained in 5 of the cases, while partial response was observed in one. However, our cases were asymptomatic and therefore we considered follow up without any treatment and owing to the fact that it is a rare disease, we aimed to present the cases and to review literature.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/;Concept: Dursun Türkmen, Kısmet Kaya; Design: Dursun Türkmen; Control/Supervision: Dursun Türkmen; Data Collection and/or Processing: Dursun Türkmen, Özlem Sezer; Analysis and/or Interpretation: Dursun Türkmen; Literature Review: Dursun Türkmen; Writing the Article: Dursun Türkmen; Critical Review: Dursun Türkmen; References and Fundings: Dursun Türkmen; Materials: Dursun Türkmen.

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