Aquagenic Palmar Wrinkling: Case Report and Literature Review

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Abstract

Aquagenic Palmar Wrinkling is a rare, transient disorder induced by brief contact with water. It has been noted in cystic fibrosis patients. We describe a unique case of a 17-year-old male with no history of cystic fibrosis, who presented with shriveled palms and multiple whitish papules following brief water immersion.

We present a 17-year-old male with an eighteen-month history of severe palm and finger wrinkling following brief (less than 5 minutes) immersion in water. The hands assume normal appearance within 60 minutes. Narrative is provided primarily by his mother as the patient has high-functioning autism. He denies associated burning or itching and is on no oral medications. Personal and family history are negative for atopic dermatitis, Raynaud's disease, and cystic fibrosis. At consultation his hands are normal (Figure 1). A photograph shown during the visit (Figure 2) revealed markedly shriveled fingers and palms and a multitude of 1mm whitish papules.

Rare cases have been reported since the first description of aquagenic palmar wrinkling (APW) by English and McCullough in 1966.[1] Subsequently, various reports have described the condition as transient aquagenic hyperwrinkling, aquagenic palmoplantar keratoderma, and transient reactive papulotranslucent acrokeratoderma.[2,3] As APW often resolves within a short period of time it is not believed to be a true keratoderma.[4]

APW has a predilection for affecting young adult women.[5,6] It is diagnosed clinically based on characteristic findings of the disease. These include the transient rapid development of yellow, white edematous papules and plaques, along with excessive wrinkling and even desquamation after brief contact with water.[7] The findings are pathognomonic and referred to as the "hand-in-the-bucket" sign.[8] Additional symptoms of pain, pruritis, and burning sensations localized to the palms are also frequently associated with the condition.

In APW, wrinkling occurs within 3 minutes of water immersion, in comparison to the normal physiologic wrinkling occurring roughly 11 minutes after water immersion.[9] It is commonly bilateral affecting the palmar surfaces with plantar involvement infrequently observed.[10] However, rare cases of unilateral involvement and varying aspects of the hands excluding the palms have been reported.[11,12] Resolution of symptoms occurs within 10-60 minutes of skin drying.[13] While the diagnosis is clinical, histopathological examination will reveal compact orthokeratosis, dilatation of eccrine sweat glands, and spongiosis.[14]

The exact etiology and pathogenesis remain unknown, although an autosomal recessive mode of inheritance has been suggested.[1] There are several conditions associated with APW including hyperhidrosis, atopic dermatitis, and Raynaud's phenomenon. The most described association of disease is cystic fibrosis (CF).[2] APW is considered a highly favorable screening tool in CF patients, with reported occurrence in 40-80% of patients.[15]

CF is an autosomal recessive condition caused by mutations in the gene involved in trans-epithelial ion transport leading to high sweat sodium concentrations. One proposed mechanism behind the prevalence in CF patients is an increased salt content in sweat leading to enhanced keratin water binding capacity of the epidermal cells.[16] The water also alters membrane stability causing vasoconstriction that contributes to skin wrinkling.[17]

APW is also associated with medications including cyclooxygenase 2 inhibitors, non-steroidal anti-inflammatory drugs, gabapentin, spironolactone, and aminoglycoside antibiotics.[18,19]These drugs may alter cell-membrane water channels, known as aquaporins, within the epidermis.[17] Additionally, the association with atopic disease supports the hypothesis of decreased barrier function. Similar to atopy, a dysfunction in stratum corneum leads to an increased ability of the skin to absorb water.[5,16]

Several treatments for APW have been identified. Topical therapies including aluminum chloride, salicylic-acid based products, and urea-containing creams targeted at reducing the hyperkeratosis and hyperhidrosis have proven of benefit.[6,20] Other therapeutic options include oral antihistamines and botulinum toxin injections. One case improved following topical treatment with the calcineurin inhibitor, tacrolimus.[13] Many cases resolve spontaneously.[21]

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