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Atypical Multiple Aquagenic Syringeal Acrokeratoderma in a Patient Treatment with Botulinum Injection: A Case Report

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Abstract

Aquagenic Syringeal Acrokeratoderma (ASA), a rare transient disease that occurs after water immersion and disappears shortly after drying, is more commonly observed in young women, which mostly affects the palms, and to a lesser extent the feet. We reported a 40-year-old Asian man with 13-year history of erythema and pale brown flattened papules on hands with no involvement of palms which was diagnosed as ASA. Additionally, knees, elbows, buttocks, and ankles were involved in the presentation of dark red macules and patches. Uniquely, the lesions were worsened and durable than usual when the patient felt exhausted. Only botulinum toxin *via* subcutaneous injection with a concentration of 50 UI per 1 ml led to significant improvement. In addition, Dermoscopy was useful and utilized for diagnosis and follow-up observation.

Keywords: Aquagenic syringeal acrokeratoderma; Botulinum injection; Dermoscopy

Introduction

Aquagenic Syringeal Acrokeratoderma (ASA), a rare transient disease that occurs after water immersion and disappears shortly after drying, is more commonly observed in young women, which mostly affects the palms, and to a lesser extent the feet [1].

Case Presentation

We reported a 40-year-old Asian man with 13-year history of erythema and pale brown flattened papules on hands with no involvement of palms (Figure 1A). Additionally, knees, elbows, buttocks, and ankles were involved in the presentation of dark red macules and patches (Figure 1E-1G). He presented with exaggerated wrinkling and eruption of white or translucent papules and plaques of lesions after their immersion in water within 10 min (Figure 1C). Sometimes the white lesions could be peeled off. It was worsened in summer, remission in winter, and faster subsiding in drier environments. Interestingly, the lesions were more severe and durable when the patient felt exhausted. Sometimes wearing sterile gloves for a long period of time can also worsen the lesions. A history of ichthyosis on the extended side of the lower legs in his childhood. Family history was significant for his father and elder sister with ichthyosis and dry skin. Further questioning revealed no history of hyperhidrosis, thyroid disease, medication use, trauma, or atopic diseases. The typical "hands-in-the-bucket" sign [2], which is not clearly visible until hands are exposed to water, was significant (Figure1 C). Dermatoscopic examination revealed scattered gravel-like keratinized flat papules, milky white and translucent, partially fused into patches, and part of whitish papules with central depression (Figure 1B, 1D). The result of the sweat chloride test was within the normal range. Therefore, the patient received a diagnosis of ASA.

Treatment with a barrier cream, 20% aluminum chloride hexahydrate, 20% urea creams, salicylic acid preparations, and Traditional Chinese Medicine was unsuccessful. The decision was made to administer Botulinum Toxin (BoNT) through infiltration [3]. One-milliliter syringe and 33G 4 mm needles were used for subcutaneous injection. Fifty units of BoNT were injected into each lesion (concentration: 50 UI per 1 ml). The injections were administered with a distance of approximately 1 cm between them. BoNT injection resulted in significant improvement in the patient the following week. There was no recurrence within three months. Dermatoscopic examination was normal before or after the patient was exposed to water.

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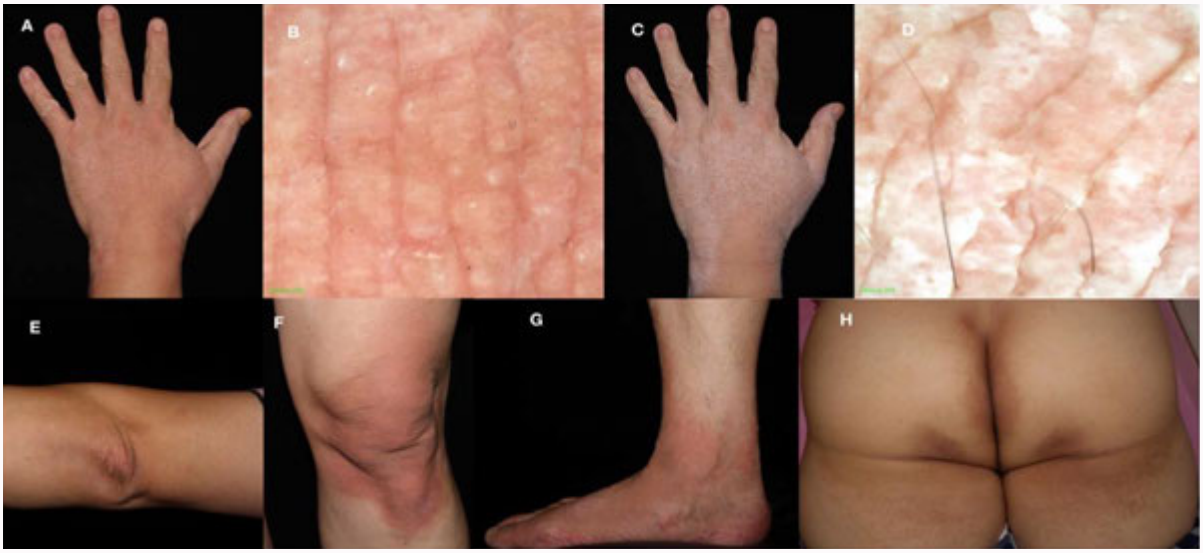

Figure 1: Aquagenic syringeal acrokeratoderma lesions. (A) Erythema and pale brown flattened papules on hands with involvement of opisthenars, dorsum of fingers, and wrist flexors. (B) Dermatoscopic examination revealed a yellow-white background with mild scales (Original magnifications ×20). (C) Translucent or whitish papules and plaques emerged companion with a mild degree of maceration after immersing hands in water for 5 min. (D) Dermatoscopic examination revealed scattered gravel-like keratinized flat papules, milky white and translucent, partially fused into patches. Part of whitish papules with central depression. (Original magnifications ×20). The atypical lesions of aquagenic syringeal acrokeratoderma with the involvement of (E) elbows, (F) knees, (G) ankles, and (H) buttocks presented the same clinical features as hands when they were exposed to water.

Discussion

In 1996, English and McCollough described 2 sisters characterized by a transient and recurrent keratoderma exclusively on the palms after water contact [4]. ASA appears to predominantly affect young females, but rare cases in males have been reported. The clinical features of ASA become obvious when patients are exposed to warm or hot water, and edematous with a thickened, translucent to whitish, pebbly appearance that mostly affects the palms can be observed [5]. Atypical presentations were occasionally reported, featuring either dorsal hands or exclusive involvement of the feet. Involvement of other sites is extremely rare. The lesions may accompany a range of symptoms such as itching, tightness, pain, or burning. The diagnosis is primarily based on clinical, and the "hands in the bucket" test is necessary. Though the exact etiopathogenesis of ASA remains unclear, sweat-duct abnormalities, hyperkeratosis, impaired stratum corneum barrier, hyperhidrosis, and elevated sweat salt concentration are considered important [6]. In addition, cystic fibrosis also has been proposed to be linked with ASA. In this case, the lesions were worse and more durable than usual when the patient felt exhausted, which may be an important basis to enrich the potential triggering factors. It is crucial to differentiate between ASA, Aquagenic Urticaria (AU), and Aquagenic Pruritus (AP) clinically. AU, a rare physical urticaria, presented with the development of itching sensation with wheal and erythema when patients are exposed to any temperature of water [7]. These lesions typically disappear within 1 h. Dermographism is negative while water challenge is positive. AP is characterized by intense itching upon water contact without visible skin lesions [8].

In our experience, the patient had no drug intake or sweat chloride test abnormality, yet BoNT injection had a significant effect, suggesting possible sweat gland abnormalities. Though dermoscopy is potentially useful in assessing and following ASA patients, only a few sporadic reports about dermoscopic features have been published. In this research, dermoscopy was utilized for diagnosis and follow-up observation.

The various treatment modalities include 20% aluminum chloride hexahydrate, 20% urea creams, aluminum lactate cream, Vaseline mixtures with 5% salicylic acid, iontophoresis, and botulinum injection [9]. Mostly, the response to treatment is unsatisfactory. Although the treatment of BoNT for ASA is limited and there is currently no unified standard for the injection dosage of BoNT, according to our case, BoNT subcutaneous injection with the concentration of 50 UI per 1 ml and the injections should be spaced approximately 1 cm apart from each other may exert a potential role in an ASA patient with atypical multiple locations.

Conclusion

We present an atypical multiple locations case of ASA in a clinical doctor whose lesions were worse and more durable under fatigue, only treated with BoNT injection that was effective, and dermatoscopy provides significant assistance in diagnosis and follow-up observation.

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