

Keratosis Pilaris Associated with Wheat Protein Allergy in an Adolescent: A Case Report

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Abstract

Keratosis pilaris is a common dermatological condition in adolescents, which usually presents because of poor cosmetic appearance of the skin. Although an exact etiology is not known, it is believed to be a multifactorial disorder associated with other medical conditions, including atopic dermatitis. However, the exact role of allergy in pathophysiology of keratosis pilaris is not known. We report an adolescent who presented with keratosis pilaris with no other allergic disorder in present or in past. The patient demonstrated presence of specific IgE to wheat protein in serum and showed complete resolution of skin lesions after wheat protein elimination. After a few weeks, accidental ingestion of snack made of refined wheat flour resulted in reappearance of lesions of keratosis pilaris within 24 hours (but not immediately), confirming the diagnosis of wheat protein allergy and suggesting mixed IgE-T cell-mediated allergy as the most likely pathophysiologic mechanism.

Keywords: Wheat Protein Allergy; Food Allergy; Mixed IgE-T Cell-Mediated Allergy; Keratosis Pilaris; Follicular Keratosis; Atopy

Introduction

Keratosis pilaris is a common dermatological disorder in adolescents. It is believed to be a disorder of keratinization but the exact etiology is not known and it is considered to be a multifactorial disorder associated with several other medical conditions. Atopic dermatitis is a common comorbidity and sometimes a differential diagnosis of keratosis pilaris, but role of allergy in keratosis pilaris is unclear. We report an adolescent female with keratosis pilaris with no allergic disorder in present or in past, who demonstrated presence of specific IgE to wheat protein, responded to wheat protein elimination, and relapsed after consumption of wheat product.

Case Report

A 12-year-old girl was brought to a pediatric allergist by her parents because she had persistent "gooseflesh appearance" of face and all four extremities for last one year. She was diagnosed with keratosis pilaris by multiple dermatologists and had been prescribed several combinations of emollients, topical corticosteroids, and oral antihistamines without any reduction in symptoms till date. She was very disturbed due to the cosmetic problem, to the extent of avoidance of going to school and frequent weeping over the last year. The parents thought of allergy as a possible cause of the disorder, which made them visit a pediatric allergist.

Her clinical history revealed that the onset of "gooseflesh appearance" of face and extremities was before one year, which was preceded by rough texture of the skin for a few months. The eruptions were occasionally associated with mild itching. No food or environmental triggers were identified for the lesions. There was no history of any allergic disorder such as allergic rhinitis, childhood asthma, atopic dermatitis, urticaria-angioedema, or food allergy. There was no history of any chronic or recurrent gastrointestinal symptoms. There was no family history of allergy or keratosis pilaris. Her physical examination showed multiple, small, goose bump-like folliculocentric keratotic papules with perifollicular erythema over face and proximal and distal parts of all four extremities. There was no eczematous lesion, xerosis, pityriasis alba, or ichthyosis. Flexural or extensor aspects of joints, axilla, groin, trunk, and scalp were spared. Rest of the physical examination was unremarkable.

Diagnosis, treatment and prognosis of keratosis pilaris were explained to the parents and it was discussed that allergy is not a likely cause of the condition. However, the parents still insisted on allergy evaluation, even if there is limited possibility of association of the condition with allergy, because they were very much concerned about possible adverse psychological consequences for their daughter, especially because she had not responded to any of the dermatological treatments till date.

Based on her diet history, wheat, milk, and chickpea were identified as most relevant food allergens in her diet and serum levels of total IgE and specific IgE for the three allergens were ordered. Her serum total IgE was 2663 UI/ml. Serum levels of specific IgE (ImmunoCAP) for milk protein, wheat protein, and chickpea protein were 0.11 kUA/L, 0.18 kUA/L, and 0.08 kUA/L, respectively.

As a next step in allergy diagnosis, wheat protein elimination for six weeks was advised. On follow up after six weeks, the parents reported significant improvement with clearance of more than half of the skin lesions, which was also evident on physical examination. After further six weeks of continued wheat protein elimination, the patient presented with normal skin with clearance of all the lesions. Based on the elimination test, possible role of wheat protein in her symptoms was suspected and she was advised to continue wheat protein elimination diet.

After three months, the patient presented with re-appearance of keratosis pilaris lesions over face and all extremities for two days. History revealed that she had accidentally consumed a commercial snack made of refined wheat flour approximately 18 hours before onset of the skin lesions. Reappearance of skin lesions with the "accidental oral food challenge" confirmed association of wheat protein allergy with keratosis pilaris in the patient.

Discussion

Keratosis pilaris is a common dermatological condition, which is especially common in adolescent population. It is characterized by keratinous plugs in follicular orifices and some authorities consider it as a normal variant [1,2]. Most patients are asymptomatic and usually the reason to visit a doctor is poor cosmetic appearance, although some patients may complain of pain or itching. Clinically, the condition presents with bumpy skin with "goose-bumps"-like appearance, also known as "chicken skin". Physical examination shows very small (one to two millimeters in size), keratotic papules centered on small hair follicles with varying degrees of surrounding erythema. Extensor surfaces of proximal upper and lower extremities and buttocks are most commonly involved; involvement of face, trunk, and distal extremities may also be seen. Although clinically benign, the condition can be associated with significant psychological distress [1].

Despite being a common condition, the exact etiology and pathophysiology of keratosis pilaris are not clear. Genetic factors have been suggested based on autosomal dominant mode of inheritance, mutations in filaggrin, and abnormalities of Ras signaling cascade [1]. Atopic dermatitis has been recognized as the most common association of the condition [1,2]; other associated conditions include ichthyosis vulgaris, obesity, scarring alopecia, ectodermal dysplasia, cardio-fascio-cutaneous syndrome, and Down syndrome. Despite association with atopy, it is not known whether food allergens play a role in pathophysiology of keratosis pilaris. Interestingly, atopic dermatitis is also an important differential diagnosis of keratosis pilaris [1,2].

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Food allergy is defined as a clinical and immunological reaction to a food allergen and is broadly classified into IgE-mediated, mixed (IgE-T cell-mediated) and non-IgE-mediated types [3]. Wheat is a common food allergen and contains more than ten allergenic proteins [4]. Wheat proteins are known to cause allergy by all three mechanisms mentioned above and to cause respiratory, gastrointestinal, and dermatologic manifestations. Among dermatologic manifestations, IgE-mediated wheat protein allergy can present with urticaria and angioedema, IgE-T cell-mediated wheat protein allergy can present with atopic dermatitis, while dermatitis herpetiformis and celiac disease are examples of non-IgE-mediated wheat protein allergy [4,5]. Keratosis pilaris is identified as one of the atypical dermatological manifestations of celiac disease and in one study, keratosis pilaris was identified as the second most common dermatologic manifestation of celiac disease in childhood and adolescence; xerosis being the most common one [6,7].

Skin prick tests and *in vitro* methods are useful to detect the presence of IgE antibodies against specific allergen. However, these tests can detect only sensitization and cannot confirm allergy. In a patient with suspected food allergy, elimination diet (elimination of suspected food allergen from diet for a specific duration) helps by both providing diagnostic information as well as by improving the patient's symptoms. After a successful elimination diet, challenge with the suspected food allergen can confirm diagnosis of food allergy [3].

In the patient discussed in the present case, the patient has keratosis pilaris without any allergic disorder, including atopic dermatitis, in present or in past. We considered a possibility of atypical presentation of atopic dermatitis, but the patient did not meet any of the UK Working Party criteria or the Hanifin and Rajka criteria - the two most popular and validated criteria for diagnosis of atopic dermatitis [8,9]. Repeated diagnosis of keratosis pilaris by multiple dermatologists and a complete lack of response to topical corticosteroids, emollients and oral antihistamines also suggested that our patient did not have an atypical presentation of atopic dermatitis.

Role of wheat protein was suspected because of detection of specific IgE to wheat protein in the patient's serum. Elimination of wheat protein in diet resulted in complete resolution of lesions of keratosis pilaris in the patient and accidental consumption of wheat protein resulted in reappearance of the lesions within 24 hours. Presence of wheat protein-specific IgE and appearance of symptoms within 24 hours (but not immediately or within six hours) of ingestion of wheat suggest mixed IgE-T cell-mediated mechanism as the most likely mechanism of food allergy in the patient. Keratosis pilaris as a manifestation of mixed IgE-T cell-mediated wheat protein allergy has not been reported in literature till date.

Conclusion

Keratosis pilaris is a common dermatological condition in adolescents and wheat is a common food allergen. Association between keratosis pilaris and food allergy is unclear. We report a case of an adolescent with keratosis pilaris as a manifestation of mixed IgE-T cellmediated wheat protein allergy.

Conflict of Interest

None.

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