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CASE REPORT

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Primary acquired cold urticaria in a 26-month-old girl: A rare case from early childhood

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Abstract

Cold urticaria is a rare type of physical urticaria that occurs upon exposure to cold stimuli. In this case report, a 26-month-old girl who developed redness, swelling, itching, and purplish discoloration on her hands, face, and feet after exposure to cold air was evaluated. Physical examination and laboratory tests revealed no pathological findings. The ice cube test was positive, and a diagnosis of primary acquired cold urticaria was made. The patient was advised on preventive measures and started on oral antihistamine therapy. During follow-up, an improvement in symptoms was observed. This case highlights that cold urticaria can also occur in early childhood and should be considered in the differential diagnosis.

Keywords: Cold urticaria, pediatric case, allergic skin reactions

Introduction

Cold urticaria (CU) is a form of physical urticaria triggered by exposure to cold stimuli such as air, water, or objects. It can present with erythema, swelling, pruritus, and in severe cases, anaphylaxis. CU can be categorized into acquired and familial types. Acquired cold urticaria (ACU) is more prevalent and includes primary and secondary subtypes, with more than 90% of patients falling under the primary group [1]. The estimated incidence of CU is approximately 0.05% in the general population, and among all physical urticarias, its prevalence ranges from 5.2% to 33.8%, depending on geographic and environmental factors [1-3]. Although the precise pathophysiology remains unclear, IgE-mediated mast cell degranulation and release of histamine and other proinflammatory mediators such as leukotrienes and platelet-activating factors are believed to play a role [1,4].

Case Presentation

A previously healthy 26-month-old girl presented with a one-month history of erythematous, pruritic, and raised lesions affecting her face, hands, and feet. The symptoms consistently occurred within five minutes of exposure to cold air and resolved when the patient returned to a warmer environment. No symptoms were noted during warm weather or contact with lukewarm water. The patient had no prior history of chronic illness, allergy, or medication use, and there was no family history of similar symptoms.

On examination, the patient was afebrile with a temperature of 36.8°C, a heart rate of 105 bpm, and blood pressure of 102/76 mmHg. Her growth parameters were within the 50th to 75th percentiles. Dermatological findings were normal at rest, and dermographism was absent.

Laboratory investigations revealed: hemoglobin 11.7 g/dL, platelets 368,000/mm3, eosinophils 110/mm3 (1.6%), ESR 12 mm/h, CRP 2 mg/L, total IgE 18 IU/mL, complement levels C3 1.01 g/L, C4 0.274 g/L, and C1 esterase inhibitor 0.375 g/L. Thyroid function tests were within normal limits (TSH 1.74 mIU/L, T4 0.96 ng/dL). Infectious disease markers, autoimmune panels (ANA, anti-dsDNA, ANCA, HIV, syphilis), and allergen-specific skin prick tests were all negative. No abnormalities were found in urinalysis or biochemical tests.

CITATION
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An ice cube test was conducted by applying an ice cube to the volar forearm for five minutes. Swelling, erythema, and pruritus appeared at the site five minutes post-removal, confirming a positive test result (figure-1). Based on clinical history and the positive cold stimulation test, a diagnosis of primary acquired cold urticaria was made. Preventive strategies, including cold exposure avoidance, were discussed with the patient's caregivers. Oral non-sedating H1-antihistamine therapy was initiated. During follow-up, significant improvement in symptoms was observed. Due to the absence of systemic symptoms or anaphylaxis, an epinephrine auto-injector was not prescribed. Informed consent was obtained from the patient's guardians for publication. A signed consent form was obtained from the patient on 19.04.2025.

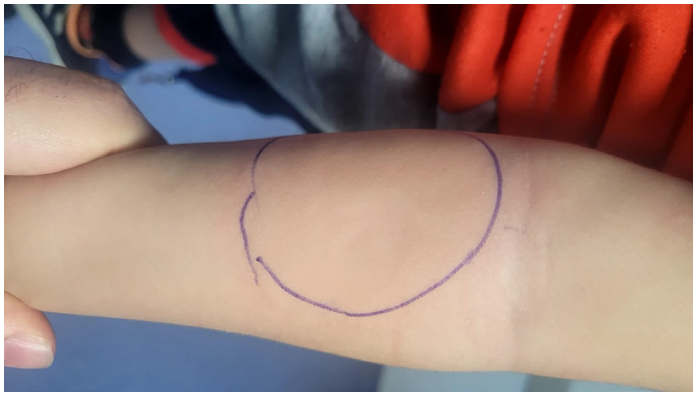

Figure 1. Induration on the forearm following the ice cube test

Discussion

ACU typically manifests with erythema, burning, and urticarial plaques within minutes of cold exposure, although delayed reactions have been reported. Lesions are usually localized but can become generalized or systemic with extensive exposure. Life-threatening anaphylaxis can occur, especially after swimming in cold water or consuming cold beverages, which may trigger oropharyngeal edema [1,5–7].

CU is rare in children, with only about 20% of cases presenting in the pediatric population. Prosty et al. reported a median age of 9.5 years in 52 pediatric CU cases, while Deza et al. and Yee et al. observed median ages of 9.1 and 8.4 years, respectively [5,8,9]. Yee et al. also documented a 4-month-old as the youngest reported case. Our patient, at 26 months, is among the youngest with confirmed primary ACU, underscoring the need to consider CU even in early childhood.

Primary ACU is idiopathic, while secondary ACU may result from insect stings, food allergies, infections (e.g., hepatitis, HIV, EBV, syphilis), vasculitis, cryoglobulinemia, and malignancies such as leukemia and lymphosarcoma [2,9,10]. In this case, no signs of infection, autoimmune disease, or malignancy were found, supporting a diagnosis of idiopathic primary ACU.

Up to 50% of CU patients have concurrent atopic conditions [4]. Yee et al. found that atopic children with CU had a higher incidence of anaphylaxis [9]. Additionally, 25% may have other physical urticarias, such as dermographism or cholinergic urticaria [4,9]. Our patient had no personal or family history of atopy, and skin prick tests and other physical stimulation tests were negative.

Diagnosis is confirmed using the ice cube test. A positive result involves wheal formation after localized cold exposure and rewarming [2,3,9–11]. If no reaction occurs, the test is repeated on the contralateral forearm. This test was positive in our case.

Differential diagnoses for CU include familial cold autoinflammatory syndrome, cryopyrin-associated periodic syndrome, paroxysmal cold hemoglobinuria, and cold-induced cholinergic urticaria. Familial types often feature delayed onset (24–48 hours) and systemic symptoms such as fever, fatigue, and elevated inflammatory markers [1,10]. These features were absent in our patient, making familial CU unlikely.

Management begins with cold avoidance and patient education. First-line pharmacologic treatment includes second-generation H1-antihistamines. If symptoms persist, antihistamine doses may be increased up to fourfold [11]. Refractory cases may benefit from biologic therapies such as omalizumab, etanercept, or anakinra [3,9,11]. For patients with a history of anaphylaxis, an emergency kit including an epinephrine auto-injector should be provided. Our patient responded well to standard-dose antihistamines, with no need for escalation or epinephrine.

Conclusion

This report presents a rare case of primary acquired cold urticaria in a 26-month-old child. The case emphasizes the importance of considering CU in the differential diagnosis of urticaria, even in early childhood. Early recognition and management, including avoidance strategies and antihistamine therapy, are essential to prevent severe reactions. Further https://orcid.org/0000-0002-1188-0918 studies are warranted to better understand pediatric CU and improve clinical outcomes.

Conflict of Interests
The authors declare that there is no conflict of interest in the study.

Financial Disclosure
The authors declare that they have received no financial support for the study.

Patient Informed Consent
Informed consent was obtained from the patient's guardians for publication. A signed consent form was obtained from the patient on 19.04.2025.

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