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CASE REPORT

Short-Term Relief with Upadacitinib: A Case Report of Palmoplantar Pustulosis in a Pediatric Patient

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Abstract: Palmoplantar pustulosis (PPP) is a chronic inflammatory skin disease with recurrent erythema, scales, and sterile pustules on the palms and soles. Treating PPP in children is difficult due to limited effective therapies, especially as there's a lack of data on upadacitinib use in children under 12.This article reports a pediatric PPP case. The patient had severe, recurring rashes that affected daily activities. Treated with upadacitinib for two months, she showed a significant therapeutic response.

Keywords: palmoplantar pustulosis, children, treatment, upadacitinib

Introduction

Palmoplantar pustulosis (PPP) stands as a persistent inflammatory dermatological condition characterized by recurrent erythema, scales, and sterile pustules manifesting on the palms and soles.[1] Presently, the availability of quality treatment regimens for PPP remains relatively limited. Recent case studies have indicated the potential efficacy of pharmacological interventions such as JAK inhibitors, particularly in ameliorating the symptoms of adult PPP,[2,3] albeit with associated risks of adverse reactions. Upadacitinib, a novel small molecule JAK inhibitor, has exhibited promising results in enhancing the clinical status of adult PPP patients.[4–6] In this case report, we present the clinical scenario of a pediatric PPP patient treated with upadacitinib for a duration of two months, resulting in a notable and substantial therapeutic response.

Case Reports

An 8 - year - old female was diagnosed with PPP at 5 due to sterile pustules on her hands. Initial treatment with hormonal ointment like calcipotriol brought temporary relief. But over three years, the pustules recurred and worsened, involving the palms, causing erythema, desquamation, and nail problems, severely affecting her daily life.

When she visited the outpatient clinic, prominent pustules, erythema, desquamation, skin thickening, nail damage, and distal onycholysis were present (Figure 1a and c). Her initial scores were PPPASI 28.8, NAPSI 39, and WI - NRS 8 (Figure 2).

Two months ago, upadacitinib (15mg once daily) monotherapy started. Within a week, PPP began to improve, with less desquamation and no new pustules. After 2 weeks, palmar pustules and nail lesions eased, with PPPASI down to 10, NAPSI to 34, and WI - NRS to 0. By 8 weeks, hand symptoms improved greatly (PPPASI 2.4, NAPSI 27), a 96% reduction from the start. Treatment stopped after 8 weeks, and by 12 weeks, skin and nail conditions continued to get better (Figure 1b and d).

Received: 8 October 2024
Accepted: 14 March 2025
Published: 3 April 2025

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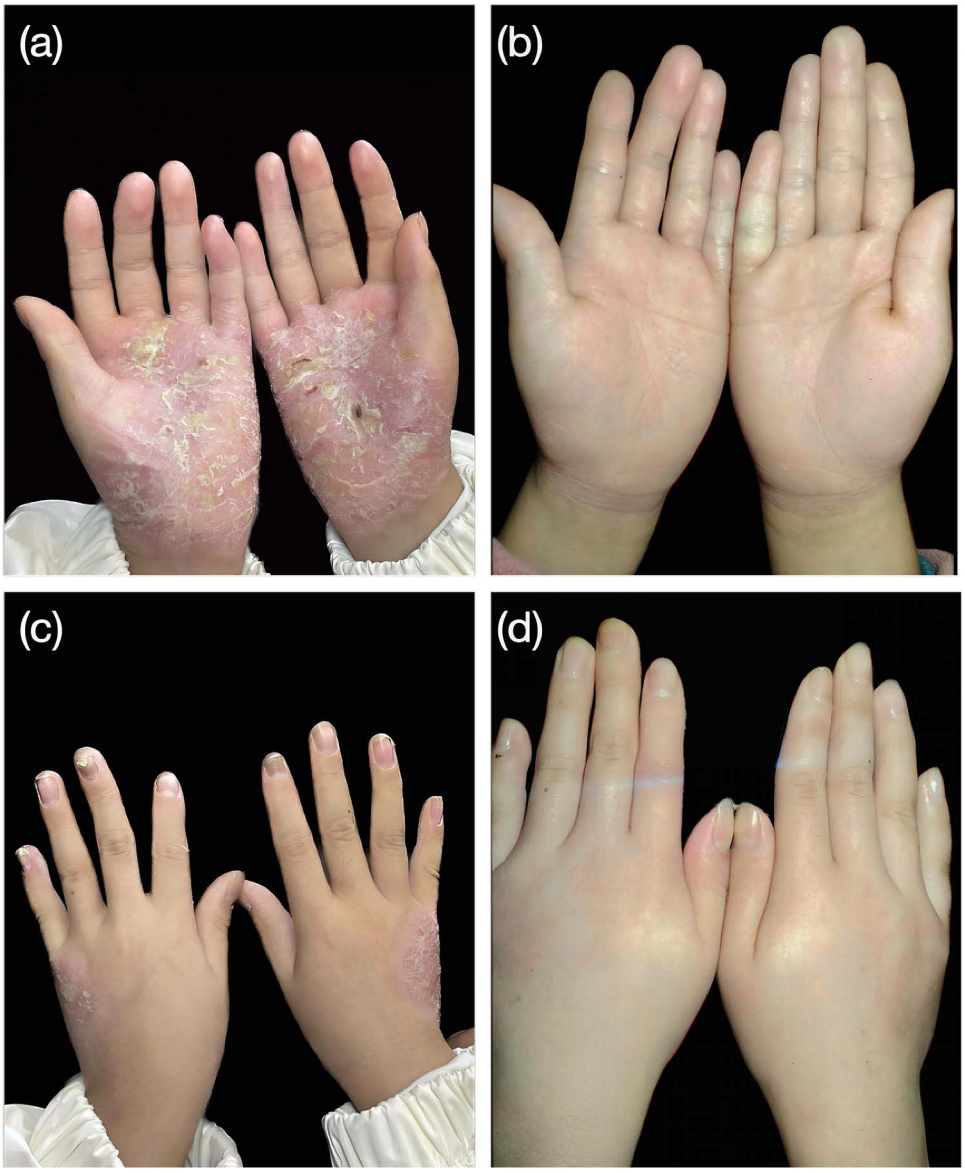

Figure 1 Clinical manifestations of PPP patients treated with upadacitinib. (**a** and **c**) are skin lesions on the patient's hands when first diagnosed. The clinical manifestations are pustules on both hands, accompanied by large areas of erythema and desquamation, skin thickening, and combined with nail damage and distal nail edge separation. (**b** and **d**) represent the clinical manifestations of PPP patients after 12 weeks: erythema and desquamation on the hands are significantly reduced, and there are no new symptoms. Pustules and nail damage have also been greatly improved.

Discussion

In this study, we present a case demonstrating the efficacy of upadacitinib in improving palmoplantar pustulosis (PPP) in a pediatric patient. The management of PPP in children poses a challenge due to the limited availability of effective treatment modalities. Existing literature contains scarce reports on the use of upadacitinib for PPP, with no prior documentation of its application in pediatric cases. The therapeutic approach to PPP remains complex, compounded by the absence of standardized guidelines or consensus on optimal management strategies. Notably, therapeutic options for pediatric PPP are notably sparse, with a dearth of pharmaceutical interventions specifically tailored for this patient population.[4,7–9]

Upadacitinib, a Janus kinase (JAK) inhibitor, was approved for medical use in both the United States and the European Union in 2019 and has been used off label as well to treat several conditions since then. It has shown efficacy in various conditions in adults, including psoriatic arthritis, atopic dermatitis, refractory ankylosing spondylitis, Crohn's disease, ulcerative colitis, refractory rheumatoid arthritis, non - radiographic axial spondyloarthritis, and PPP. Clinical

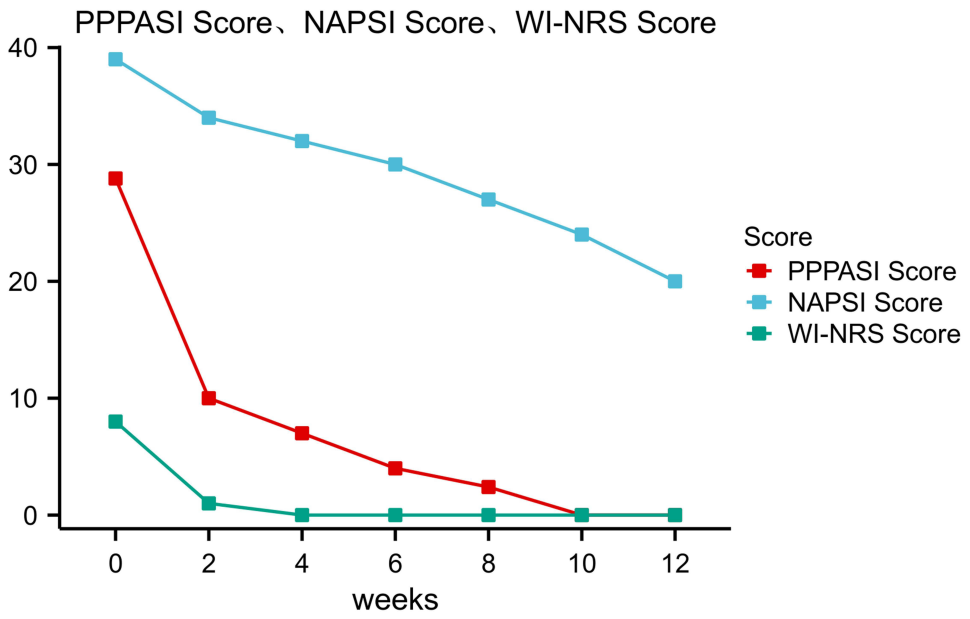

Figure 2 Palmoplantar Psoriasis Area and Severity Index (PPPASI), Nail Psoriasis Severity Index(NAPSI) and Worst Itch Numeric Rating Scale (WI-NRS) scores for evaluating the therapeutic efficacy of upadacitinib. The abscissa represents the treatment time (unit: weeks), the red line represents the PPPASI score, the blue line represents the NAPSI score, and the green line represents the WI-NRS score.

investigations involving minor patients have predominantly focused on adolescents within the age range of 12 to 18 years. Limited reports exist regarding the effectiveness and safety of upadacitinib in treating atopic dermatitis in adolescent populations.[10–12] Notably, there is a lack of case studies and safety data on upadacitinib use in children under 12 years old. While safety data for upadacitinib indicates favorable tolerability and comparable efficacy and safety profiles to those observed in adults[10–12] further clinical trials are imperative to ascertain the safety and efficacy of upadacitinib specifically in children under 12 years of age.

We suggest that the short-term use of upadacitinib may have a rapid and beneficial effect on children with PPP. Specifically, a short-term course of upadacitinib followed by discontinuation after improvement could be a promising treatment approach for severe pediatric PPP. However, further clinical studies are essential to validate the outcomes observed in this case and to establish the efficacy and safety of this treatment strategy in pediatric patients with PPP.

Data Sharing Statement

The data that support the findings of this study are available from the corresponding author upon reasonable request.

Ethics Statement

Ethics approval: This work was approved by the Medical Ethics Committee of Fangshan Hospital, Beijing University of Chinese Medicine (Approval No. FZJ JS-2021-002). All participants provided written informed consent prior to enrollment. In the case of the minor patient described in this manuscript, written informed consent for the publication of the case details and any associated images was obtained from the patient's parent. The consent form clearly explained the purpose, potential uses, and possible risks related to the publication of the case information. The identity of the patient has been protected to the greatest extent possible in the manuscript to ensure confidentiality.

Acknowledgments

All authors have approved the submission of this manuscript. This manuscript has not been published or submitted elsewhere.

Funding

This work was supported by the Zhejiang Provincial Key Laboratory of Traditional Chinese Medicine Cultivation for Arthritis Diagnosis and Treatment (grant number:C-2023-W2083).

Disclosure

The authors declare no conflicts of interest in this work.

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