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CASE REPORT

Idiopathic plantar hidradenitis in a 5-year-old girl after exposure to wet footwear

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Idiopathic plantar hidradenitis (IPH) is characterised by tender erythematous papules and nodules which involve the soles of the feet. The pathogenesis of this condition remains obscure, with a temporal association between exposure to cold and moisture and the presentation of skin lesions which may allude to a possible pathogenic mechanism.

We report a five-year-old girl who developed plantar hidradenitis. The patient participated in a school outing wherein her feet were exposed to prolonged cold and damp. She complained of pain in the soles of her feet and was unable to walk the next morning. She was admitted to hospital with a suspicion of infection and discharged the next morning on oral antibiotics. Review by dermatology raised the possibility of plantar hidradenitis and a course of low dose oral steroids was given. The patient recovered fully in five days.

Familiarity with the inciting tiggers and symptoms of IPH in children allows a clinical diagnosis to be made without the need for hospitalization and further investigations. The authors believe that the instigating factors in our patient were prolonged exposure to wet footwear combined with strenuous activity. Further studies are needed to determine the aetiology of IPH.

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Idiopathic plantar hidradenitis (IPH) is characterised by tender, erythematous papules and nodules which involve the soles of the feet. This condition tends to affect younger children.[1] It is also known as 'traumatic plantar urticaria', 'palmoplantar eccrine hidradenitis' and 'plantar erythema nodosum'.[2–4] IPH is histologically similar to neutrophilic eccrine hidradenitis (NEH), with notable exceptions which include the presence of neutrophilic abscesses localized to the eccrine coils within the deep reticular dermis and the lack of syringosquamous metaplasia.[5,6] The pathogenesis of this condition remains obscure but there is a temporal relationship between exposure to cold and moisture and the presentation of skin lesions which may allude to a possible pathogenetic mechanism.[1] This case report aims to increase the awareness of this condition as a potential differential diagnosis in order to avoid unnecessary hospital admissions, histological investigations and treatments such as antibiotics.

CASE REPORT

We report the case of a healthy five-year-old girl who presented with bilateral erythematous, tender nodules after she participated in a school outing wherein children visited a valley and jumped in muddy puddles in the morning causing her shoes to become wet. On returning to school, her schoolteachers changed her into dry socks, but the child remained in her wet shoes until returning home around 6 hours later. Her behaviour was normal, and she did not complain of any pain. Her parents bathed and dried her thoroughly.

The child spent the rest of the day playing on a scooter and running around outside. She complained of some pain in her feet in the evening which her parents attributed to her eventful and physically demanding day. The patient slept through the night but, on waking up she, complained of bilateral sole pain and could not weight bear. She was therefore taken to the local emergency department.

On examination she was afebrile and systemically well, having only very tender bilateral erythematous nodules (Figure 1A) which precluded her from weight bearing. She was admitted for intravenous cefixime to cover for possible infection, given the history of exposure to potentially contaminated water. Bloods investigations included a full blood count which only showed marginally raised white cells (13.26 x10^9/L) with relative neutrophilia (8.39 x10^9/L) and a slightly raised CRP (6.2mg/L). A COVID swab and blood culture were unremarkable. She was therefore discharged with a 7-day course of cefpodoxime to cover for possible infection.

Outpatient dermatology review raised the possibility of plantar hidradenitis as the working diagnosis and introduced a course of low-dose oral steroids, 10mg prednisolone daily for 3 days. The following day, the erythematous nodules had reduced in size (Figure 1B) and the patient was on her feet with some pain and could manage walking on tip toes. She had almost completely recovered by that same evening, returned to school the next day, and even managed more strenuous activities such as tennis.

Significant improvement was seen once the course of steroids was completed (Figure 1C).

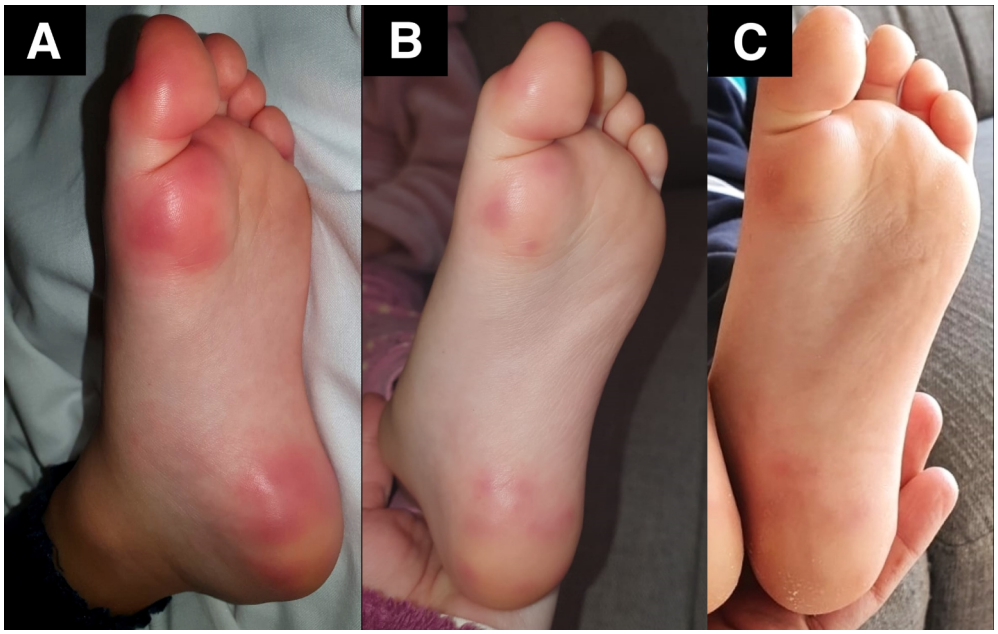

Figure 1 **A)** *On presentation before prednisolone was given. Erythematous nodules mostly present on the left heel, toes, and forefoot. B) One day post-prednisolone. Erythematous nodules on the left heel, toes, and forefoot smaller than the previous day. C) Significant improvement seen upon completion of the course of prednisolone.*

DISCUSSION

IPH is a condition characterised by the onset of erythematous and tender nodules on the plantar aspect of the foot. A variant of this condition may also affect the palms, when it is referred to as idiopathic palmoplantar hidradenitis (IPPH).[1,3] IPH was first reported in 1988 where its initial pathophysiology was thought to be related to strenuous physical activity.[2] It is now recognised that a number of inciting factors may trigger this condition.[1] These may include strenuous activity, trauma, heat, excessive moisture, perspiration and pseudomonas infection. The aetiology of this disease is uncertain although an association with intense physical activity and pseudomonas infection is under investigation.[7]

Reports of recurrent palmoplantar hidradenitis have also been made. This is a benign condition which may also be associated with low-grade fever. Resolution occurs in the absence of therapy making the disease self-limiting. It has been suggested that this entity may be more common than is reported in literature but is not routinely evaluated histologically given its transient nature.[8]

Our case fits within the typical clinical presentation of idiopathic plantar hidradenitis. The history of prolonged exposure to wet footwear combined with strenuous activity is concordant with other case reports of the same condition.[1,3]

The differential diagnosis of this condition may include erythema nodosum, Sweet syndrome, Behçet's disease, chilblains, Neutrophilic eccrine hidradenitis (NEH), Acute acral eruptions (AAE) secondary to SARS-CoV-2 infection and Pseudomonas hot-foot sysndrome.[6,9–11] The latter has also been described as one of the possible contributing factors of the condition. This case report discusses each of the listed differentials in the context of our case and how these were ruled out given the clinical picture.

Erythema nodosum is a cutaneous reaction consisting of inflammatory, tender, nodular lesions. The pathology may be associated with a plethora of diseases including infections, autoimmune disorders, inflammatory bowel diseases, sarcoidosis, rheumatologic diseases, medications and malignancy.[12] Our case involved a healthy child with none of the mentioned co-morbidities; erythema nodosum was therefore not likely. IPH should be distinguished from palmoplantar erythema nodosum as both are clinically characterized by tender erythematous nodules which appear after physical activity and that may involve the palms and/or soles of children.[12]

Sweet syndrome (SS), also known as acute febrile neutrophilic dermatosis and Behçet's disease (BD) form part of a heterogeneous group of inflammatory skin disorders known as neutrophilic dermatoses. BD typically presents in patients in their third to fourth decades while SS typically presents in patients between 47 and 57 years of age. Although both may present with the abrupt onset of painful erythematous nodules or plaques with predominantly neutrophilic infiltrates in the dermis, neither condition fits within the clinical picture of our case.[13]

Chilblains often appear in the differential diagnosis of IPH.[14] Chilblains usually resolve spontaneously and involve localized inflammation of the skin which occurs upon exposure to cold, wet weather. The inflammation is a result of a maladaptive vascular response to non-freezing cold.[15] The child in our case was exposed to cold, wet conditions however, dermatological input did not feel that the lesions were typical of chilblains and a course of steroids resulted in complete resolution. A clinical diagnosis of IPH was therefore made.

The clinical picture of IPH contrasts with that of with neutrophilic eccrine hidradenitis (NEH) which typically occurs in patients receiving chemotherapy for haematological malignancy. The clinical features of NEH are polymorphic and can affect other areas besides the palms and soles. While both clinical and histological investigations are needed to make a diagnosis of NEC, histological investigation is not required to make a diagnosis of PH.[8,16] Moreover, NEH typically resolves after cessation of chemotherapeutic treatment and the application of topical steroids.[10]

COVID-19 remains pandemic at the time of writing. Acute acral eruptions (AAE) secondary to SARS-CoV-2 infection have been observed in children and the clinical picture is similar that of IPH. AAE presents with heterogeneous features which include erythematous and violaceous papules and macules.[11] The patient in our case was asymptomatic and tested negative for COVID-19 on the day of admission. AAE was therefore not likely.

Pseudomonas aeruginosa is a recognized cause of folliculitis which arises after the use of swimming pools, hot tubs and contact with other bodies of water. Presenting features may include bilateral vesicular, pustular, or maculopapular lesions and pruritic folliculitis. Conversely, pseudomonas hot-

foot syndrome is a benign, self-limiting condition, the aetiology of which seems to involve *P. aeruginosa* infection. This condition, however, does not require antibiotic therapy.[9] The patient in our case was exposed to potentially contaminated water and thus, potentially, to P. aeruginosa. The authors do not exclude that the microbe may have contributed to the pathophysiology in this case as there have been documented reports of IPH triggered by *P. aeruginosa* infection.[7,9]

Familiarity with the inciting triggers and symptoms of IPH in children allows a clinical diagnosis to be made without the need for hospitalization and further investigations such as biopsy. Analgesic treatment, rest and topical or systemic steroid therapy seem to be the only effective treatment options. Biopsy to demonstrate eccrine gland neutrophil infiltration should only be carried out in abnormally prolonged cases or cases where there is an atypical presentation.[16,17] Increasing awareness of this condition may help avoid unnecessary investigations and treatments such as courses of antibiotics, especially given that the differential diagnosis often includes infective conditions such as pseudomonas hot-foot syndrome.[9]

The authors believe that the instigating factors in our patient were prolonged exposure to wet footwear combined with strenuous activity which induced trauma to the soles of the feet. We do not, however, exclude that *P. aeruginosa* exposure or infection may have contributed to the pathophysiology in this case.

LEARNING POINTS

1. Familiarity with the inciting triggers and symptoms of IPH allows a clinical diagnosis to be made without the need for hospitalization and further investigations.
2. Analgesic treatment, rest and topical or systemic steroid therapy seem to be the only effective treatment options.
3. Biopsy to demonstrate eccrine gland neutrophil infiltration should only be carried out in abnormally prolonged cases or in cases with atypical presentation.

CONCLUSION

Familiarity with the inciting triggers and symptoms of IPH in children may help avoid hospital admission, unnecessary investigations, and treatments, such as antibiotics. The importance of the multidisciplinary approach is highlighted in this case as the final diagnosis was only postulated once the patient had been seen by a general practitioner, paediatricians, and a dermatologist. Input from all members of the caring team and involving a specialist dermatologist allowed a clinical diagnosis to be made, appropriate treatment to be instituted and inappropriate treatment to be terminated. Further studies are needed to determine the aetiology of IPH and perhaps to allow more targeted treatment.

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