

BRIEF ARTICLE

A Case of Pseudoverrucous Papules and Nodules in an Adult with Spinal Dysraphism

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ABSTRACT

Pseudoverrucous papules and nodules (PPN) occur in the setting of chronic irritant contact dermatitis commonly due to urine or feces. PPN most commonly affects infants but has also been reported in association with urostomies, colostomies, chronic urinary incontinence, encopresis, chronic fecal incontinence, spinal cord injury, and potentially spinal dysraphism. Herein, we present the case of a patient with spina bifida in association with Dubowitz syndrome who presented with perianal PPN. To our knowledge, there is only one other case reported in the literature of a patient with spina bifida who presented with PPN.

INTRODUCTION

Pseudoverrucous papules and nodules (PPN), an uncommon entity, occurs as a result of chronic irritant contact dermatitis from repeated exposure to urine and feces. PPN most commonly occurs in infants but has also been reported in adults in association with urostomies, colostomies, chronic urinary incontinence, chronic fecal incontinence, and potentially spinal dysraphism.¹ Dubowitz syndrome is a rare autosomal recessive disorder characterized by microcephaly, developmental and motoric delay, failure to thrive, abnormal facies, and short stature. Less than 200 cases have been reported in literature and life expectancy is often decreased in most cases.⁵ Herein we present a case of perianal PPN in a patient with Dubowitz syndrome and spina bifida.

CASE REPORT

A 42-year-old male with a history of Dubowitz syndrome and associated spina bifida resulting in chronic urinary and fecal incontinence presented to the dermatology clinic with a perianal rash of an unknown duration. Of note, the patient denied any sensory perception in the area due to his spinal dysraphism. On physical exam, a verrucous, macerated plaque with surrounding hyperpigmentation was noted along his inferior sacrum and superior intergluteal cleft (**Figure 1**). The differential diagnosis at this time included condyloma acuminata, verrucous carcinoma, squamous cell carcinoma, and pseudoverrucous papule and nodules (PPN) secondary to chronic irritant contact dermatitis. Bacterial and fungal cultures were obtained (both negative) as well as a punch biopsy for histopathology.

Punch biopsy revealed pseudoepitheliomatous verrucous epidermal hyperplasia and underlying dilated vessels with papillary edema. The histopathological findings taken together with the clinical presentation were consistent with PPN. The patient was started on a combination of triamcinolone 0.1% ointment and nystatin 100,000 units/gram twice daily in addition to frequent diaper changes with liberal use of a zinc oxide-containing barrier cream. Significant clinical improvement was observed 2 months after initiation of treatment (**Figure 2**).



Figure 1. Multiple verrucous nodules coalesced to form plaques at the perianal area.

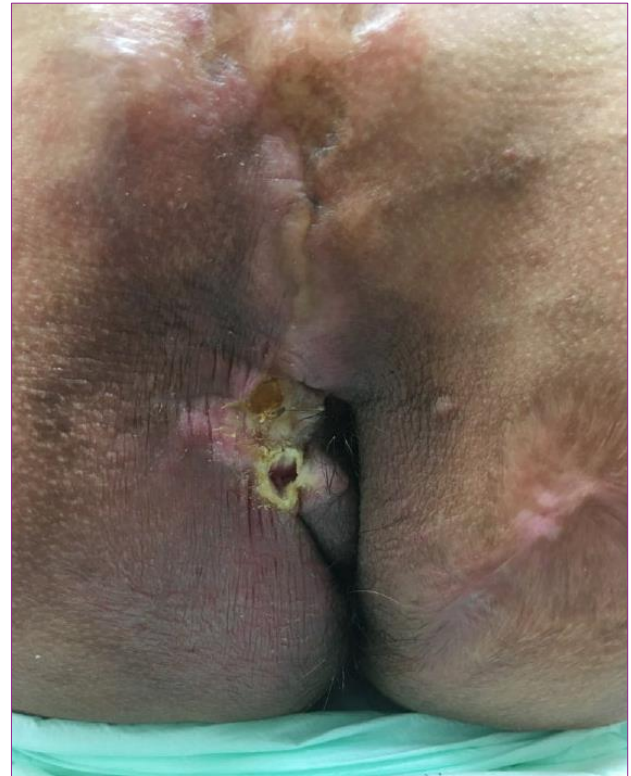


Figure 2. Macerated plaque at the perianal area with minimal overlying crust.

DISCUSSION

PPN is a rare clinical manifestation of chronic irritant contact dermatitis associated with repeated exposure to urine or feces. It typically presents as multiple, gray or erythematous, dome-shaped or verrucous papules, plaques, and nodules in genital, perianal, and peristomal sites.¹ Histopathologically, this condition demonstrates marked psoriasiform epidermal hyperplasia with broad hyperparakeratosis.¹ A moist environment and increase in pH secondary to ammonia present in feces and urine increases the permeability of the skin and compromises its natural barrier function.² Most cases of PPN in the literature are described in children and in patients with ostomies. To our knowledge, there is only one other case in the literature,

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reported by Dandale et al., of PPN in an adolescent patient with spina bifida.¹

The mainstay of therapy for PPN focuses on the management of fecal/urinary incontinence and resulting irritant contact dermatitis. If incontinence cannot be corrected, meticulous hygiene practices should be implemented including frequent diaper changes with gentle cleansing and drying of the area followed by liberal use of barrier creams such as petroleum or zinc to protect the skin from irritation by urine and feces.⁴ Also, vinegar soaks and oral ascorbic acid can be used to acidify the urine and reduce irritation.³ Secondary infections are a relatively common complication of PPN and the area should be monitored closely for signs of infection.

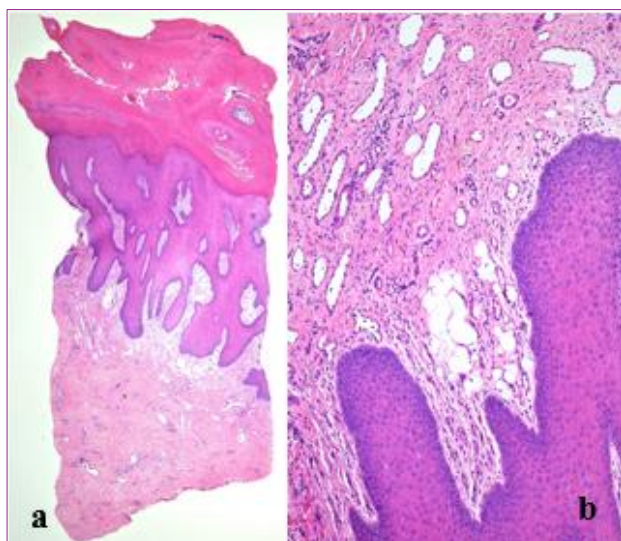


Figure 3. a) 20x low power photomicrograph: Pseudoepitheliomatous verrucous epidermal hyperplasia with marked hyperparakeratosis and underlying dilated vessels with papillary dermal edema consistent with pseudoverrucous papules (lymphedema). **b)** 100x high power photomicrograph: Acanthotic epidermis with bulbous irregular downgrowths and associated increased ectatic dermal vessels with papillary dermal edema.

CONCLUSION

In conclusion, pseudoverrucous papules and nodules (PPN) is an uncommon condition that can clinically mimic several infectious, inflammatory, and neoplastic processes but should be suspected in cases of chronic irritant contact dermatitis from urine and feces. Furthermore, this case demonstrates the rare association between PPN and spinal dysraphism.

Conflict of Interest Disclosures: None

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