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Single Case

Recurrent Granuloma Gluteale Infantum Secondary to Fecal Overflow Incontinence

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Abstract
Granuloma gluteale infantum is a rare pediatric dermatological disorder of uncertain etiology. Suggested causes include fluorinated corticosteroids, *Candida albicans*, and irritant contact dermatitis. We present the case of a 3-year-old boy with recurrent episodes of granuloma gluteale infantum which resolved with treatment of his fecal overflow incontinence. As each recurrence correlated with a relapse of overflow incontinence, in this case the cause was irritant contact dermatitis from the liquid stool. This is the first reported case of recurrent granuloma gluteale infantum.

Introduction

Granuloma gluteale infantum (GGI) is a chronic inflammatory skin disease that most commonly occurs during infancy. Grossly, the lesions appear as violaceous papules and nodules in the gluteal region. The disease was originally named "vegetating bromidism" in 1891 due to its occurrence with the application of ointments containing bromide [1]. It was later named granuloma gluteale infantum by Tappeiner and Pfleger [2] in 1971. While the etiology of GGI

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is uncertain, possible causes cited in the literature have included irritant contact dermatitis, infection with *Candida albicans*, and previous treatment with fluorinated steroids. Treatment is usually conservative, involving barrier creams and eliminating sources of irritation. The process typically resolves within several weeks [3]. We present a unique case of GGI that started shortly after birth and persisted over several years. After diagnosis, it was successfully treated and had multiple recurrences. As each recurrence correlated with a relapse of overflow incontinence, the GGI was secondary to irritant contact dermatitis from the liquid stool.

Case Presentation

A 3-year-old male Caucasian presented with the complaint of "persistent sores" that had been present since shortly after birth. The child was 7 weeks premature and at discharge from NICU was noted, according to his mother, to have sores in the diaper area. His pediatrician diagnosed the eruption as "diaper dermatitis." The child was treated with multiple over-the-counter preparations including zinc oxide, antibacterials, and antifungals. His mother denied using topical corticosteroids. Record review showed no prescription of topical corticosteroids. The problem persisted through the first year of life with defecation becoming progressively painful, and continuous passage of loose stool. He was referred to a pediatric gastroenterologist for fecal soiling. A radiograph showed fecal material throughout the colon. He was diagnosed with fecal overflow incontinence and treated with laxatives. Nodular skin changes were noted by the gastroenterologist but were not connected to leakage of stool. These changes were noted 1 year prior to dermatologic referral. His symptoms did not improve with treatment and his mother did not take him for follow-up. At the time he was seen, defecation, cleaning, and sitting in a car seat were difficult because of severe pain.

Physical exam revealed approximately 30 red, moist granulomatous papules that ranged in size from 2 mm to 1 cm in the perianal region (Fig. 1a). Some of the lesions were confluent, forming plaques. Many of the lesions were eroded and friable. After being cleaned for examination, the patient was incontinent with liquid stool 5 min later (Fig. 1b).

A biopsy showed pseudo-verrucous epithelial hyperplasia with areas of spongiosis, parakeratosis, and overlying crust (Fig. 2a). There was papillary dermal edema and a mixed superficial infiltrate consisting of lymphocytes, plasma cells, neutrophils, and scattered eosinophils (Fig. 2b).

Treatment with enemas and topical zinc oxide was initially unsuccessful as the child actively resisted treatment from the mother because of pain. After additional education on the primary role of bowel care for resolution of overflow incontinence and support from the grandmother, treatment with laxatives, enemas, and creams was initiated. Two weeks later the eruption had cleared (Fig. 1c, d). The regimen of oral bulk agents, periodic enemas, and topical zinc oxide was continued.

The child experienced several recurrences of GGI. Each of the recurrences occurred during custody with the father who favored natural treatments and disagreed with laxatives and enemas. There was a recurrence of overflow incontinence and GGI with stopping bowel care. Reinstitution of bowel and skin care resulted in rapid GGI clearance. Direct communication with the child's pediatrician ultimately convinced the father to continue bowel care with complete resolution of GGI.

Discussion

GGI is a rare manifestation of diaper dermatitis with less than 30 reported cases [4]. It occurs during infancy with violaceous nodules appearing in the diaper area [5]. The etiology is not well understood but it is thought to be a result of chronic irritation with urine and feces leading to inflammation and maceration. Infection with *C. albicans* and use of fluorinated steroids have also been implicated [5, 6]. While the cause is typically uncertain, in this case the cause was irritant contact dermatitis. When the overflow incontinence recurred, the eruption also recurred due to continual irritation from liquid stool. This is the first reported case of recurrent GGI.

The general characteristics of the primary lesion of GGI are oval, firm to hard, red to purple colored nodules. The lesions align along the long axis parallel to skin lines. Size can range from 5 mm to 4 cm. The surface can be smooth, lichenified, or eroded/ulcerated depending on the stage of lesions. The lesions have been described to persist up to 6 weeks and spontaneously regress over 2–4 weeks if the precipitating agent is removed. Postinflammatory hyperpigmentation and atrophic scars can be observed as the lesions resolve [3].

GGI histology typically shows parakeratosis, acanthosis, spongiosis, and exocytosis. There is predominantly a mixed perivascular inflammatory dermal cell infiltrate [3, 5, 6]. Ortonne et al. [7] looked at the lesions ultra-structurally and found the presence of intracytoplasmic structures resembling rickettsia-like bodies within dermal macrophages.

Some authors have suggested that GGI is on a disease spectrum with perianal pseudoverrucous papules and nodules (PPPN) and Jacquet's dermatitis [8]. Others argue that GGI is its own severe variant of diaper contact dermatitis [9]. PPPN is a disease that can occur in adults around urostomies and colostomies and in children when there is chronic irritation with stool. Clinically, the two conditions may appear very similar. Histologically, they differ in that GGI has a dense dermal infiltrate that is typically absent in PPPN [10]. A similar variant occurs in adults called granuloma gluteale adultorum. It can occur in bedridden or movement-restricted adults. Histologically it appears identical to GGI [11]. Robson et al. [8] reviewed the subject and found that granuloma gluteal infantum, PPPN and Jacquet's dermatitis were more similar then dissimilar. They suggested that the multiple names were confusing and that these disorders should be grouped as erosive papulonodular dermatosis [8].

The exact pathophysiology of GGI has yet to be elucidated. In most instances, there is a history of a precipitating inflammatory skin conditions such as seborrheic dermatitis, *Candida* infection, or irritant contact dermatitis [5, 6]. It is thought to be due to inflammation, maceration, and secondary infection. Items that come in contact with the skin such as commercial diapers, laundry detergents, baby wipes, topical halogenated corticosteroids, urine, and fecal material have all been implicated as contributing factors [12].

Treatments that have been used successfully include barrier products, topical and intralesional corticosteroids, and oral loperamide [12–15]. Flurandrenolide-impregnated tape was shown to produce drastic improvements in 2 treated patients [13]. However, the use of steroids is controversial as one report found an increase in the size of lesions after steroid use [14]. Rapid improvement of erosive papulonodular dermatosis can occur if the source of skin irritation can be removed or mitigated [15]. Our patient experienced rapid resolution and rapid recurrence that corresponded with exposure to liquid stool. One commonly used product for mitigating contact dermatitis is using barrier treatment with zinc oxide. Zinc oxide is a physical protectant that is safe, inexpensive, and readily available over the counter [12]. The primary goals of therapy are centered on avoiding irritation, reducing patient discomfort, and early intervention to prevent scarring.

In summary, clinicians should be aware that while irritant contact diaper dermatitis commonly presents as erythema, edema, and scale; incontinent children may present with nodular diaper dermatitis.

Statement of Ethics

The authors have no ethical conflicts to disclose.

Disclosure Statement

The authors declare that there is no conflict of interest regarding the publication of this paper.

References

1 Simmons IJ. Granuloma gluteale infantum. Australas J Dermatol. 1977 Apr;18(1):20–4.
2 Tappeiner J, Pfleger L. [Granuloma gluteale infantum]. Hautarzt. 1971 Sep;22(9):383–8. German.
3 Sweidan NA, Salman SM, Kibbi AG, Zaynoun ST. Skin nodules over the diaper area. Granuloma gluteale infantum. Arch Dermatol. 1989 Dec;125(12):1703–4.
4 Krol AL, Krafchik BR. Diaper area eruptions. In: Eichenfield LF, Frieden IJ, Mather EF, Zaenglein AL, editors. Neonatal and infant dermatology. 3rd ed. London: Elsevier Saunders; 2015. p. 245–64.
5 Al-Faraidy NA, Al-Natour SH. A forgotten complication of diaper dermatitis: granuloma gluteale infantum. J Family Community Med. 2010 May;17(2):107–9.
6 De Zeeuw R, Van Praag MC, Oranje AP. Granuloma gluteale infantum: a case report. Pediatr Dermatol. 2000 Mar-Apr;17(2):141–3.
7 Ortonne JP, Perrot H, Thivolet J. [Granuloma gluteale infantum: ultrastructural study (author's transl)]. Ann Dermatol Venereol. 1980 Jul;107(7):631–4. French.
8 Robson KJ, Maughan JA, Purcell SD, Petersen MJ, Haefner HK, Lowe L. Erosive papulonodular dermatosis associated with topical benzocaine: a report of two cases and evidence that granuloma gluteale, pseudoverrucous papules, and Jacquet's erosive dermatitis are a disease spectrum. J Am Acad Dermatol. 2006 Nov;55(5 Suppl):S74–80.
9 Rodríguez Cano L, García-Patos Briones V, Pedragosa Jové R, Castells Rodellas A. Perianal pseudoverrucous papules and nodules after surgery for Hirschsprung disease. J Pediatr. 1994 Dec;125(6 Pt 1):914–6.
10 Goldberg NS, Esterly NB, Rothman KF, Fallon JD, Cropley TG, Szaniawski W et al. Perianal pseudoverrucous papules and nodules in children. Arch Dermatol. 1992 Feb;128(2):240–2.
11 Fujita M, Ohno S, Danno K, Miyachi Y. Two cases of diaper area granuloma of the adult. J Dermatol. 1991 Nov;18(11):671–5.
12 Konya J, Gow E. Granuloma gluteale infantum. Australas J Dermatol. 1996 Feb;37(1):57–8.
13 Kikuchi I, Jono M. Letter: flurandrenolide-impregnated tape for granuloma gluteale infantum. Arch Dermatol. 1976 Apr;112(4):564.
14 Bluestein J, Furner BB, Phillips D. Granuloma gluteale infantum: case report and review of the literature. Pediatr Dermatol. 1990 Sep;7(3):196–8.
15 Yap FH, Thom G. Perianal pseudoverrucous papules and nodules in Hirschsprung's disease: rapid resolution with oral loperamide. Pediatr Dermatol. 2017 Nov;34(6):e343–4.

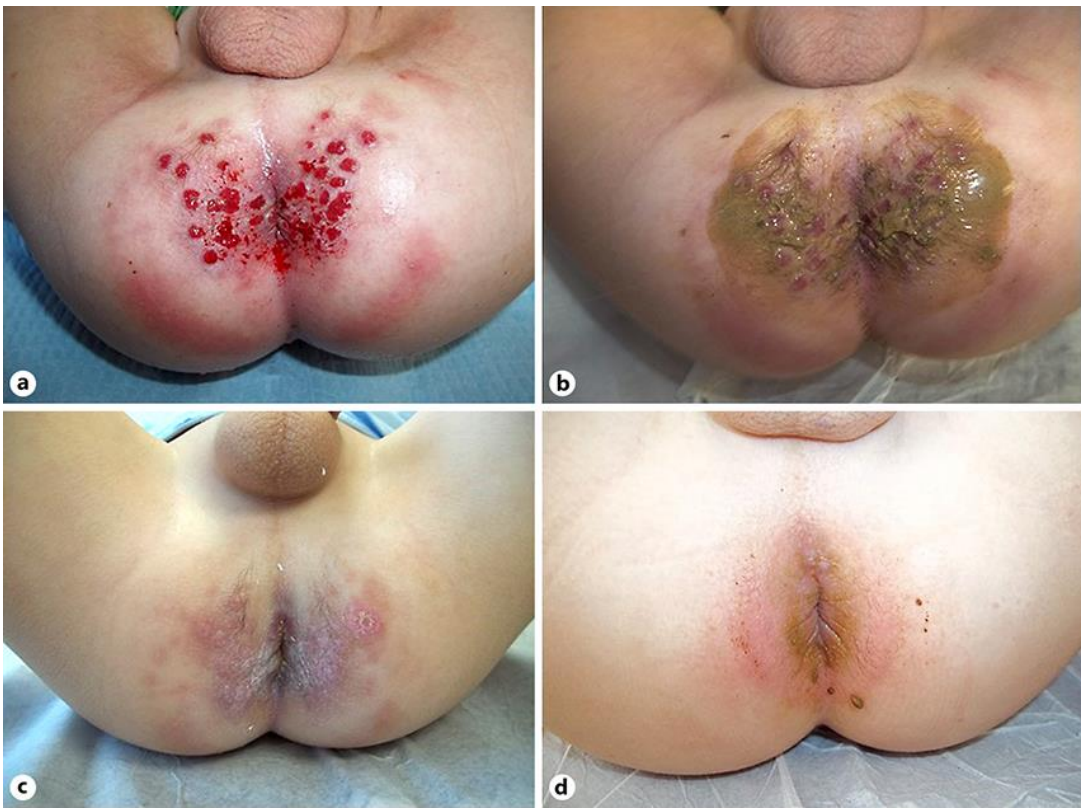

Fig. 1. **a** Initial presentation after cleaning showed numerous red, friable papules and nodules surrounding the anus. **b** 5 min after cleaning, the patient was incontinent with liquid stool. **c** Two weeks after treatment with barrier creams and enemas. **d** Three months after continuous treatment.

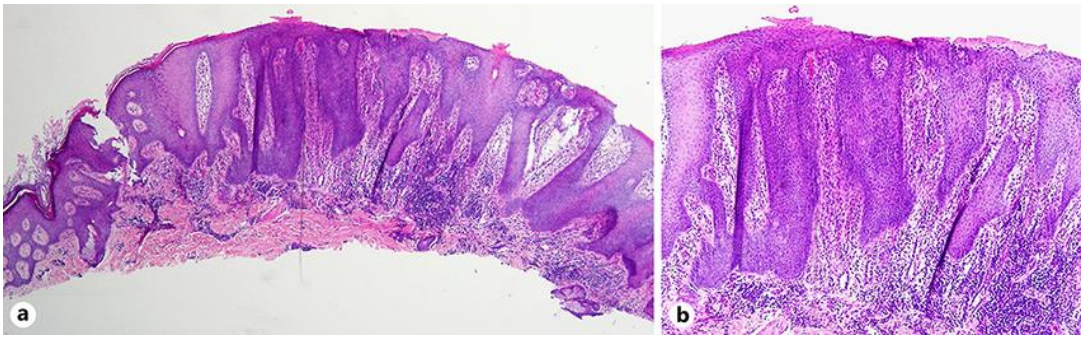

Fig. 2. **a**, **b** There is epithelial hyperplasia with parakeratosis, spongiosis, and overlying crust. Additionally, there is papillary dermal edema and a mixed superficial infiltrate consisting of lymphocytes, plasma cells, neutrophils, and scattered eosinophils. **a** ×40. **b** ×100.