Relapsing subcutaneous nodules of the penis in a pediatric patient

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Abstract

A 14-year-old boy presented with a history of non-tender, subcutaneous coalescing nodules located on the ventral-lateral aspects of the penis shaft for one year. Laboratory investigations for blood count and autoimmunity were within normal limits. Complete excision was performed, and on histology, the dermis showed necrobiotic material composed of altered collagen bundles, surrounded by a palisade of histiocytes and scattered lymphocytes, thus allowing a diagnosis of subcutaneous granuloma annulare. Only 18 published cases reported penile granuloma annulare. Medical management was advocated in 7/18 cases, either as a first-line or adjuvant therapy where surgery was not radical. Three patients received high-potency local steroids: two cases underwent adjuvant sessions of intralesional triamcinolone, and one patient received pentoxifylline orally. Surgery should be considered a second-line option since 5/8 of treated cases eventually recurred. The pentoxifylline-treated case witnessed a relapse after drug discontinuation, while topical steroids lead to complete recovery without relapses.

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Key words: granuloma annulare, children, lump, genitals, penis.

Contributions: AS, wrote the majority of the paper; ESGDA, revised the paper critically; LN, is accountable for the accuracy of the work; MB, MML, gave a substantial contribution to the study design; MC, gave the final approval for the paper.

Conflict of interest: the authors declare no potential conflict of interest.

Patient consent for publication: the authors obtained written consent.

Availability of data and materials: data and materials are available from the corresponding author upon request.

Received: 12 February 2023.
Accepted: 12 February 2023.
Early view: 10 August 2023.

Dermatology Reports 2023; 15:9687
doi:10.4081/dr.2023.9687

Case Report

A 14-year-old Caucasian boy presented a month-history of non-tender, subcutaneous coalescing nodules located on the ventral-lateral aspects of the penis shaft. The lesions developed over one year and gradually increased in size. The mass was skin-colored, hardly moveable under the skin, firm, and lobulated on palpation (Figure 1).

There was no history of trauma, and his family history was unremarkable. The patient was otherwise healthy and presented a connective tissue nevus on his back, since childhood. Laboratory investigations for blood count and autoimmunity, including antinuclear antibodies (ANAs), extractable nuclear antigen and anti-DNA antibodies, were within normal limits.

Due to the growing evolution and the unclear diagnosis, complete excision was performed. On histology, the dermis showed necrobiotic material composed of altered collagen bundles, surrounded by a palisade of histiocytes and scattered lymphocytes. The histochemical stains of Ziehl-Neelsen, FITE, and PAS were negative for possible microorganisms (Figure 2). The histological findings finally allowed a diagnosis of subcutaneous granuloma annulare (SGA).

Discussion

Granuloma annulare (GA) is an idiopathic disorder of the dermis and hypodermis, occurring in both genders and all age groups.

Histology is characterized by palisading eosinophilic granuloma (so-called *blue* granuloma), consisting of central degeneration of collagen with mucins, surrounded by interstitial and palisade lymphohistiocytic infiltrate. It presents with 5 distinct clinical variants, namely localized (the most common), generalized, perforating, patch, and subcutaneous. SGA is most common among children and has a slight male predilection. Elective locations are the antero-tibial plateau, ankles, dorsal feet, buttocks, hands, scalp, and eyelids.[1]

Only 18 published cases reported penile GA, with the involvement of the penile shaft (13/18, 72.2%), glans (1/18, 5.6%), the base of the penis (1/18, 5.6%), foreskin (1/18, 5.6%), or a combined glans and shaft (2/18, 10.2%).[2] Among the 13 cases with

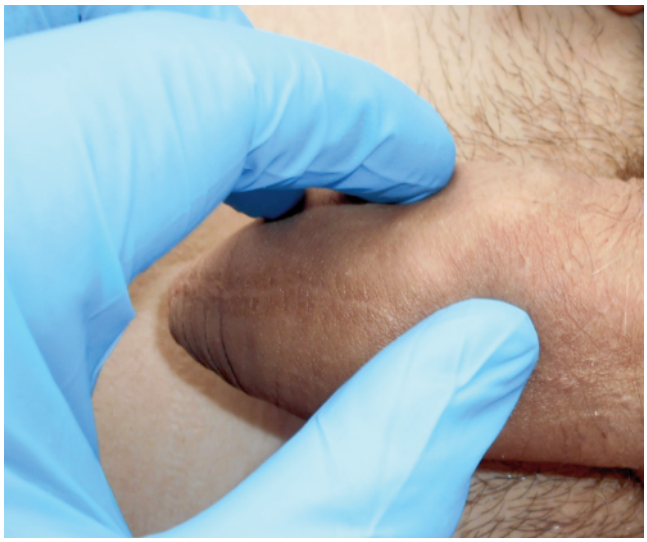

Figure 1. Clinical presentation of lobulated, fleshy, moveable subcutaneous nodules located at the penis shaft. The overlying skin was unremarkable.

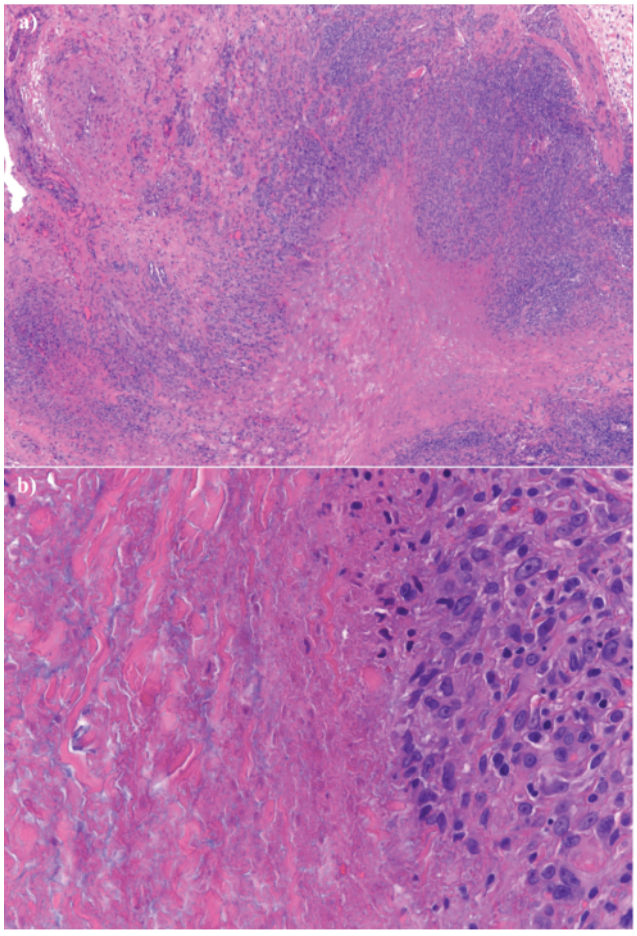

Figure 2. Hematoxylin and Eosin stain: **a**) Low-power view showing presence within the dermis of necrobiotic material surrounded by a palisade of histiocytes and scattered lymphocytes (original magnification ×50); **b**) High-power view highlighting the altered collagen bundles with intervening mucinous material and histiocytes at the periphery (original magnification ×200).

shaft involvement, 3 report a limited dorsal involvement,[3-5] while our case predominantly affects the ventral aspects.

SGA pathogenesis is still debated: both antibody-dependent cell-mediated cytotoxicity and delayed-type hypersensitivity have been postulated. Different studies documented intravascular IgM and complement deposits and activated T-cell infiltrates on sample specimens, supporting both mechanisms.[6] According to Mercieca and coworkers, which reviewed all penile GA cases, the age of onset is highly heterogeneous (range: 7-80 years), as is the disease duration (3 weeks-3 years). Pediatric GA (aged less than 14 years) accounted for 4/18 (22.2%) of cases.[5-8] 50% of patients were referred to urology and genitourinary clinics, while only 10.6% received a dermatologic consultation.[2]

Therapeutic abstention was proposed in 4/18 cases, whereas complete/partial surgical excision was pursued in 8/18 cases.[2] In one case the nodules were excised at the time of circumcision,[9] whereas the same procedure was performed for treating recurrence after excision in another patient, leading to complete resolution.[4] Medical management was advocated in 7/18 cases, either as a first-line or adjuvant therapy where surgery was not radical. 3 patients received high-potency local steroids (0.5 mg/g of clobetasol propionate or 0.1% mometasone furoate),[2] 2 cases underwent adjuvant sessions of intralesional triamcinolone,[4,6] and one patient received pentoxifylline 400 mg, 3 times a day orally.[6] Overall, surgery should be considered as a second-line option since 5/8 of treated cases eventually recurred. The pentoxifylline-treated case witnessed a relapse after drug discontinuation,[6] while topical steroids lead to complete recovery without relapses in all cases.[2]

The response rate possibly correlates with the administered treatment and may be conditioned by the anatomic SGA localization, since all relapsing cases involved the penis shaft.[2] This explains why the totality of glans SGA showed an excellent response to topical steroids.[2]

Our patient witnessed a complete resolution after the procedure. However, after a time lag of 2 months, the lesions eventually recurred at the proximal borders of the previous ones. Clobetasol propionate 0.05% ointment administered 5 days per week for 2 months led to no results. No further therapy was prescribed due to the lack of symptoms. At 1-year follow-up, the patient reported the periodic onset of new lesions at the penis shaft and penis base, and the clearance of the older ones, which last averagely 3 months.

Conclusions

SGA differential spectrum is broad, and should include infectious granulomas, rheumatoid nodules, necrobiosis lipoidica, epithelioid sarcoma, epidermoid cysts, Peyronie's disease, deep penile syringomas, and lymphatic tumors. Clinical history and examination are often decisive. However, in doubtful cases, the histological examination remains mandatory.

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