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Subcutaneous Granuloma Annulare of the Penis Associated with a Urethral Anomaly: Case Report and Review of the Literature

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Abstract: **Granuloma annulare (GA) is an uncommon benign inflammatory skin condition, most often found on the extremities of young females. The subcutaneous variant of GA involving the penis is very rare. We report a case of subcutaneous GA associated with a urethral anomaly in a 15-year-old boy that persisted for a year. Treatment options are discussed with a review of the current literature.**

Granuloma annulare (GA) is an uncommon benign inflammatory disease of the dermis, usually involving the hands and feet, predominantly diagnosed in young females. The disease is characterized by focal degeneration of collagen with a surrounding area of reactive inflammation and fibrosis (1–4). Penile GA is remarkably rare and an uncommon presentation of the disease. To the best of our knowledge, only 15 cases have been previously reported in the literature. Herein we present a new case of penile GA associated with a urethral anomaly and review the literature.

CASE REPORT

A 15-year-old boy presented to our pediatric surgery clinic with a 1-year history of asymptomatic penile nodules that progressively increased in number and size. His medical and family history was unremarkable. There was no history of trauma, allergy, diabetes, or rheumatoid arthritis, and he did not have any urinary symptoms. Routine laboratory studies were normal. Physical examination revealed multiple pea-sized subcutaneous lesions largely on the left side of the penile shaft limited to the distal third of the foreskin. The lesions were neither painful nor pruritic and overlying skin was intact (Fig. 1). The foreskin was extremely tight, with a phimotic ring. After preputial retraction, a 2-cm-long blind-ending tract was noted dorsally, running parallel with the normal urethra (Fig. 2A). At surgery, after the normally located meatus was exposed, a 1-cm dorsal-running sinus was found that did not communicate with the dorsally located tract (Fig. 2B). The urethra was otherwise normal (Fig. 2C). Circumcision was

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performed together with removal of all penile lesions and the dorsally located tract. The blind-ending sinus in the meatus was left intact because it did not interfere with normal micturition. Histologic examination revealed granulomatous lesions with necrobiotic collagen fibers surrounded by palisading histiocytes, mucin, and few giant cells, consistent with subcutaneous GA (Fig. 3). At the 3-month postoperative follow-up, he presented with a new lesion on the penile shaft (Fig. 4). Considering the benign nature of subcutaneous GA and the high risk of recurrence, the patient and the parents were reassured, and the decision was made to continue follow-up without further treatment.

DISCUSSION

GA is a benign inflammatory disease, more commonly occurring in girls, involving focal degeneration of collagen and a surrounding area of reactive inflammation and fibrosis. It can appear anywhere on the skin, but the hands and feet are frequently involved (2,5–7). There are four clinical variants (localized, generalized, subcutaneous, and perforating forms), each with a characteristic clinical manifestation (3,8). The subcutaneous form is rare and most commonly affects the skin of bony prominences, especially the anterior tibia, feet, hands, and occipital scalp of children younger than 5 (9). GA of the penis is rare,

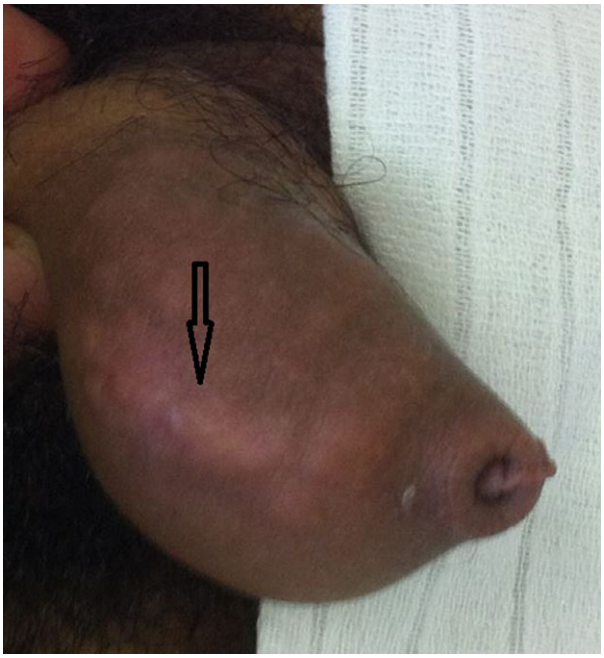
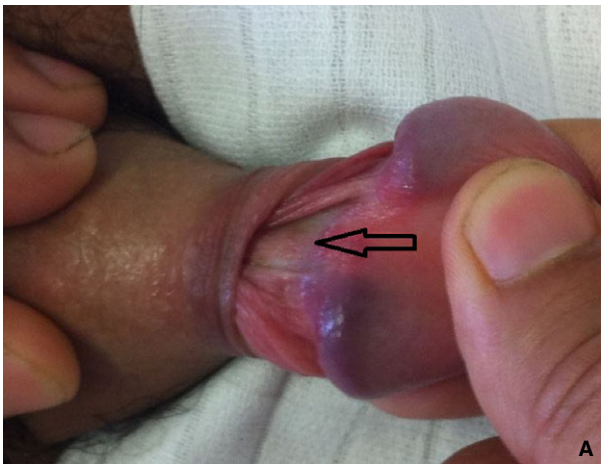
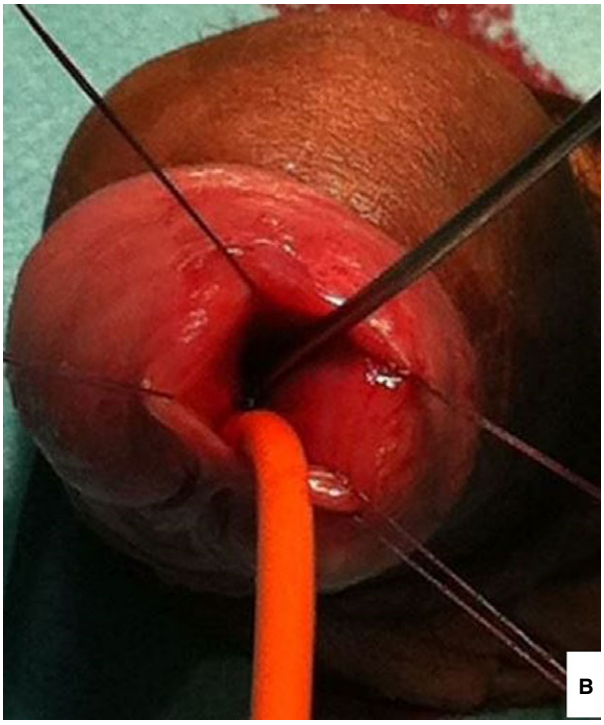


Figure 1. Multiple penile subcutaneous granuloma annular nodules of varying size.





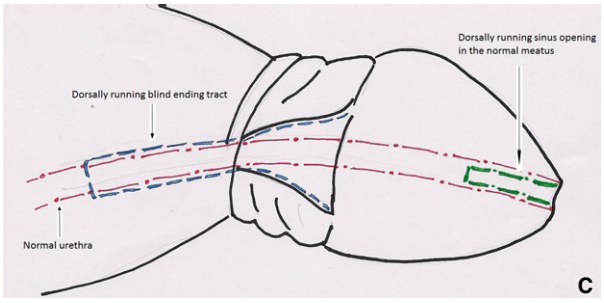


Figure 2. (**A**) Dorsally running blind-ending tract (arrow). (**B**) Probe shows 1 cm long sinus running parallel to the normal urethra, marked by a urethral catheter. (**C**) Dorsal schematic drawing of the penis showing the normal urethra, blind ending tract and the sinus opening in the meatus.

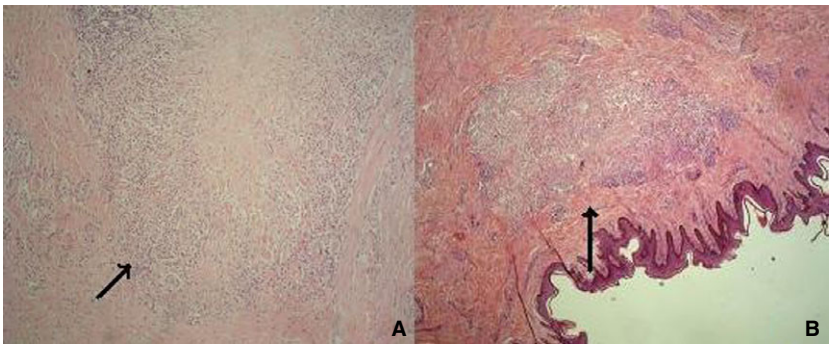


Figure 3. Histologic examination showing subcutaneous granuloma annulare. (**A**) Localized necrobiotic collagen fibers surrounded with histiocytes and rare giant cells. (**B**) Palisaded granuloma.

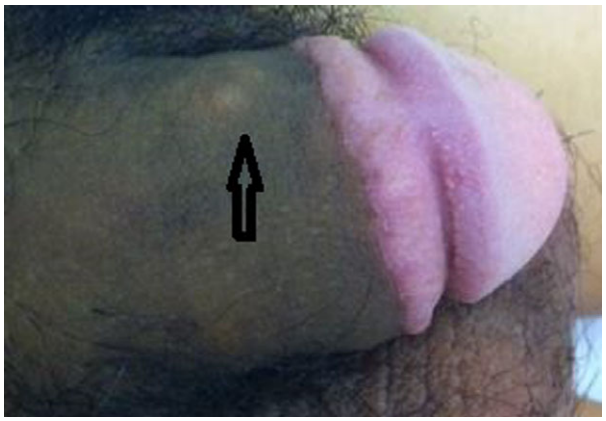


Figure 4. Recurrence of the subcutaneous granuloma annulare lesion 3 months postoperatively.

with only 15 cases previously reported in the English medical literature, 10 of which were classified as subcutaneous GA (10–14).

The etiology of GA is unknown, although a few factors, such as primary necrobiosis, trauma, and fungal, and viral infection, are proposed as contributing factors. The disease process remains unclear, although there are four as-yet-unproven theories: vasculitis leading to necrotic changes, trauma-induced necrobiosis, monocytic release of lysosomal enzymes, and type IV hypersensitivity reaction (1,8). As in our case, tight foreskin or phimosis may increase the risk of infection and trauma and contribute to the development of GA.

Histologic findings in GA include degenerated collagen surrounded by palisading inflammatory cells, which are mostly histiocytes mixed with monocytes, possibly with a few lymphocytes and fibroblasts (9,11–13,15). The differential diagnosis of penile GA includes benign neoplasms, sexually transmitted diseases, warts, and ringworm (4,14,16).

Subcutaneous GA of the penis is characterized by multiple firm, nontender nodules of varying diameters covered by normal-color skin. The age at diagnosis ranges from 7 to 61 years, but most affected men are in their third decade of life (9,11).

Suggested treatments for GA include steroids (intralesional injection, topical, or systemic) and surgical removal, which are associated with varying degrees of success. Localized trauma or diagnostic biopsy may initiate resolution of the lesions (8,17,18). Postsurgical recurrence of GA at other sites in children occurs in up to 79% of reported cases (5). Kossard et al (14) performed circumcision at the time of removal of GA nodules, and there was no recurrence 2 years after surgery. In our case, a new nodule developed 3 months after circumcision.

CONCLUSION

Localized GA is an asymptomatic, self-limiting disease and may resolve spontaneously within a few years. Penile subcutaneous GA is remarkably rare and the lesions are asymptomatic, so many patients may remain undiagnosed. Accurate histologic diagnosis and familiarity with subcutaneous GA of the penis can prevent overtreatment. Patient reassurance and a noninvasive approach is advocated, and aggressive surgical removal is unnecessary, except in selected cases related to concerns with cosmetic appearance or patient anxiety.

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