

Case – Penile verrucous epithelial hyperplasia

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Introduction

Masses of the penis are uncommon, especially in the pediatric population, and they need to be promptly assessed and biopsied. We present what we believe to be the first case of rapidly recurring glans and foreskin verrucous epithelial hyperplasia, a premalignant lesion. The young patient, a 13-year-old uncircumcised teenager, presented an itching lesion growing over four months and rapidly recurring after resection.

Case report

A 13-year-old uncircumcised teenager with a past medical history of asthma, autism, and attention deficit hyperactivity disorder, presented to our department for the evaluation of a penile lesion after initial evaluation by dermatology. The mass was first noticed on the glans 18 months prior to the consultation but was disregarded by the patient. However, he finally requested medical attention because the lesion rapidly progressed over a four-month period. Clinically, the chief complaint was pruritus without pain, discharge, or urinary symptoms.

Initial physical exam showed a 1.5 cm beige ulcerative and crateriform lesion on the right lateral side of the glans (Fig. 1). Testes were normal in size and shape, as was retraction of the foreskin for this Tanner 5 teenager. There was no palpable inguinal lymph node. Considering the absence of other risk factors, such as infection or phimosis, the dermatologist considered the lesion to be the results of chronic scratching. Topical antibiotic treatment was attempted but was not effective. Biopsy without sedation was not feasible, therefore, we performed a first surgical excision of the 1.5 x 1.0 x 0.5 cm glans tumor and of a 0.5 x 0.5 x 0.3 cm satellite tumor on the inner foreskin. The main lesion was

sent for frozen section, which showed a benign verrucous and papillomatous epithelial lesion and negative margins. The final local and revised histology reports (by pathologist at Necker-Enfants Malades Hospital, Paris) described a marked epidermal hyperplasia with numerous dyskeratotic cells scattered at different levels of this epidermis. There was no atypia nor mitotic figures or koilocytes (Fig. 2). Human papillomavirus (HPV), immunohistochemistry typing, and mycosis cultures were negative.

At one-month followup, we noted two new lesions near the previous surgical excision area (Fig. 3). Masses were removed with deeper and larger surgical margins. Macroscopic examination of the specimen showed beige masses of 0.4 x 0.3 x 0.2 cm and 1.8 x 1.3 x 0.6 cm. Histological slides showed similar findings and the Necker-Enfants Malades Hospital's pathologist concluded the diagnosis was a "penile verrucous epithelial hyperplasia."

Two months later, a new lesion appeared on the right side of the glans at great distance from the previous resection sites. Partial penectomy with glans resection and entire corpora cavernosa preservation was performed (Fig. 4). Surgical margins were clear. Histopathology showed similar findings to what was previously seen.

After 18 months of followup, the patient is well without any urinary problems. Penile sensitivity is intact without residual pain or erectile dysfunction. No shaft relapse has been seen.

Discussion

We present a patient with a very atypical clinical presentation for a penile lesion considering his young age and the absence of risk factors. He was not sexually active, a non-smoker, uncircumcised, without phimosis, and HPV tested negative. Furthermore, the lesion relapsed multiples times in a short period following surgeries, which makes this case even more unusual.

The differential diagnoses for pediatric penile lesions include infectious and sexually transmitted diseases, inflammatory conditions, traumatic causes, and pre-malignant and malignant lesions, even though these lesions remain



Fig. 1. 1.5 cm beige ulcerative and crateriform lesion on the right lateral side of the glans.

very rare in children. Cancers of the penis and pre-malignant penile lesions are considered diseases affecting older patients, mostly men aged between 50 and 70 years old.¹ The incidence in Europe and in the U.S. is approximately 1/10 000,² with incidence increasing with age and with the prevalence of HPV. Squamous cell carcinoma (SCC) is the most common pathology type. Risk factors are numerous and include untreated phimosis that induces chronic inflammation,³ HPV infection and penile trauma, phototherapy, psoriasis, smoking, poor hygiene, low social and economic status, and marital status.⁴

Based on the current literature, penile cancer has rarely been documented in the pediatric population. Penile rhabdomyosarcoma (RMS) in a three-year-old child⁵ and 16-year-old boy⁶ have been reported, with a painless mass that can be treated by surgery, chemotherapy, and radiation therapy.



Fig. 3. At one-month followup, two new lesions near the previous surgical excision area were noted.

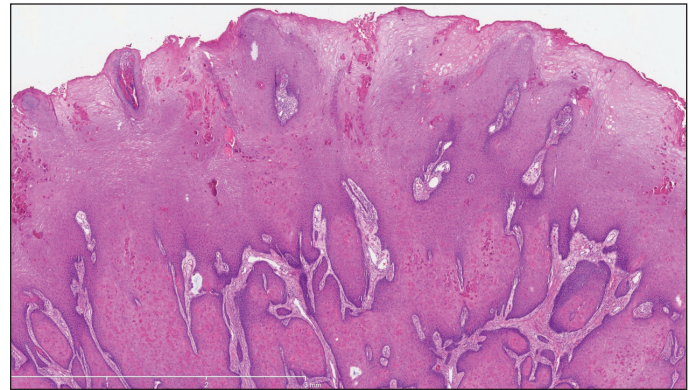


Fig. 2. Histology showed marked epidermal hyperplasia with numerous dyskeratotic cells scattered at different levels of this epidermis. There was no atypia nor mitotic figures or koilocytes.

Congenital penile teratoma⁷ and glomus tumor of the penis⁸ in pediatric patients have been registered. In our report, the final diagnosis was verrucous epithelial hyperplasia (VEH), which is usually a premalignant lesion⁹ that can transform into verrucous carcinoma (VC), a very well-differentiated variant SCC. It has been published as an oral mucosal lesion but has never been reported in genital tumors in the pediatric population.¹⁰ This type of lesion has been associated to tobacco use and HPV.

Conclusions

Although this is a unique case — the first reported in a teenager and in the genital apparatus — VEH should be considered in the differential diagnosis of a penile lesion.

We strongly believe that any penile mass should be rapidly investigated, particularly in children without risk factors,



Fig. 4. Two months later, a new lesion appeared on the right side of the glans at great distance from the previous resection sites. Partial penectomy with glans resection and entire corpora cavernosa preservation was performed.

by specialized dermatologists and urologists to provide a prompt and appropriate treatment.

Competing interests: The authors do not report any competing personal or financial interests related to this work.

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