

Buried penis: An unrecognized risk factor in the development of invasive penile cancer

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Abstract

One of the documented benefits of neonatal circumcision is protection against invasive penile cancer. To date there have been a handful of published cases of invasive penile cancer in men circumcised as neonates. We report a case of a 73-year-old man, with a history of neonatal circumcision with no evidence of previous human papillomavirus exposure, who developed a buried penis secondary to obesity. He was diagnosed with Grade 2, pT3N0 squamous cell carcinoma of the penis. This report suggests that buried penis may pose a risk factor for the development of penile cancer despite the protective effects of neonatal circumcision. Thus periodic examination of a buried penis is warranted even in patients with no risk factors for penile cancer. A review of the literature is provided.

Introduction

Penile cancer usually affects males in the sixties and seventies. Risk factors for developing the disease include the presence of foreskin,^{1,3} phimosis,^{4,6} infection with HPV and the subsequent development of warts,^{7,8} poor penile hygiene and the irritating effects of smegma,⁹ chronic balanitis or trauma,^{10,11} prior treatment with psoralen and ultraviolet A photo chemotherapy (PUVA) for psoriasis,¹² tobacco use of any kind,^{10,11,13} and high risk sexual behaviours with multiple sexual partners.¹⁰

Currently, there is a lack of literature addressing the topic of a buried penis and its risk for developing penile cancer. We report a case of invasive penile cancer in a male who underwent neonatal circumcision and then as an adult developed a buried penis secondary to aging and obesity. We propose that buried penis itself may pose a contributing risk to the development of penile cancer, despite the documented protective effects of neonatal circumcision.

On a global level, upwards of 30% of males are circumcised, making circumcision the most common surgical procedure in the world.¹⁴ On average, there are about 25 circumcisions being performed per minute worldwide.¹⁵

Most of these surgeries are performed for religious or cultural reasons and are therefore termed non-therapeutic. There are also therapeutic circumcisions in an attempt to treat an underlying pathologic process. A debate that continues to linger, and has for some time, is whether non-therapeutic circumcisions should be endorsed by health professionals. At the present time, the Canadian Pediatric Society and the American Academy of Pediatrics do not recommend routine circumcisions of newborns. Their stance stems from the fact that there are no valid medical indications for circumcision in neonates.^{16,17} Some of the documented benefits conferred by neonatal circumcision include decrease incidence of urinary tract infection (UTI) in the first year of life, decreased risk of HIV infection, decreased risk of human papillomavirus (HPV), and decreased incidence of invasive penile cancer.^{18,19} A long-standing misconception in medicine, dating back to the early 1930s, is that neonatal circumcision confers absolute protection against invasive penile cancer.²⁰

Case

A 73-year-old man presented in urinary retention to the emergency room. After failed attempts at Foley catheter insertion, the patient underwent suprapubic cystostomy to decompress the bladder. Catheter insertion failed as the patient had a buried penis and a firm, indurated mass at the glans that extended to the shaft of the penis. This lesion caused a stricture just proximal to the urethral meatus making it difficult for catheter insertion. The patient described a remote smoking history and he was circumcised as a neonate. There was no history of HPV infection or high risk sexual behaviour. There was no history of trauma to the penis and the patient was not concerned about the lesion as he was not fully aware of it because of his buried penis. The patient had recurrent UTIs in the past year and described episodes of hematuria that had resolved prior to his presentation. The patient's medical history was significant for type II diabetes mellitus, hypertension, dyslipidemia and basal cell carcinoma on the head and back treated with local excision and radiation.

On examination, the patient had a buried penis with an indurated lesion at the glans that extended to the shaft of the penis. There was no palpable lymphadenopathy, digital rectal examination and examination of the scrotum and testicles was unremarkable.

The patient went on to have a biopsy of the penile lesion under local anesthesia. The pathology came back as invasive moderately differentiated squamous cell carcinoma. Prior to surgery, he underwent computed tomography (CT) of the chest, abdomen and pelvis, which was unremarkable. A preoperative magnetic resonance imaging revealed a mass centred in the glans penis, involving distal ends of both corpora cavernosa and the corpus spongiosum – causing urethral obstruction (Fig. 1). Inguinal lymphadenopathy was not identified.

The patient underwent radical penectomy and creation of a perineal urethrostomy. The final pathology revealed a Grade 2, pT3 squamous cell carcinoma of the penis with no evidence of lymphovascular or perineural invasion (Fig. 2). Immuno-histochemical staining for p16, a surrogate marker for HPV infection, was negative. As a result of the grade and stage of the penile cancer, this patient was at risk for harbouring disease in the inguinal lymph nodes.²¹ With no palpable lymphadenopathy, the established guidelines would dictate that the patient undergo a modified bilateral radical inguinal lymph node dissection. All lymph nodes assessed ($n = 4$) were negative for malignancy and therefore conversion to the standard inguinal lymph node dissection was not indicated. The patient continues with routine surveillance and has now been disease-free for more than 24 months.

Discussion

Penile cancer, although a relatively rare disease in the Western world, is a disease with a high morbidity and mortality rates. The incidence of penile cancer varies among populations. These variations in incidence rates are related considerably to the practice of neonatal circumcision. For example, the lowest incidence of penile cancer is reported among Israeli Jews, at 0.1/100 000, where neonatal circumcision is universally practiced.²² Yet, neonatal circumcision does not confer absolute protection against the development of penile cancer. Our literature search (in PubMed with neonatal circumcision and penile cancer as keywords) found 16 published cases of penile cancer in men circumcised as neonates.^{2,23-34} Most recently, Saibishkumar and colleagues³⁴ reported three separate cases of invasive penile cancer in men circumcised as neonates. A commonality among all these patients was their remote history of HPV infection, a factor thought to play a critical role in the development of invasive penile cancer. The specific subtypes of HPV that have been implicated in the development of in situ and invasive penile cancer include 16, 18, 31 and 33.³⁵ Of these, HPV 16 is the most frequently detected subtype in primary carcinomas of the penis.³⁵ We report, for the first time, a

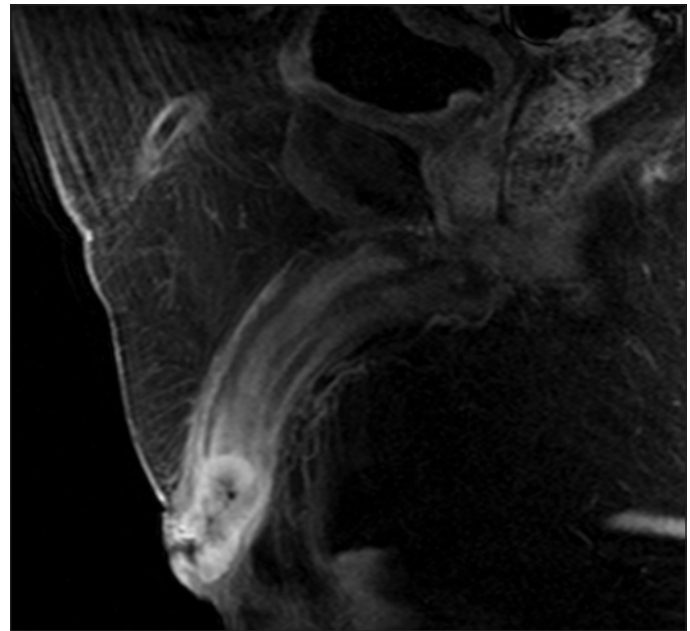


Fig. 1. A preoperative magnetic resonance imaging showing a mass centred in the glans penis causing urethral obstruction.

case of a circumcised man, with no history of HPV infection who developed invasive penile cancer in a buried penis.

Buried penis, also referred to as hidden penis, inconspicuous penis and/or concealed penis, is a condition in which the penile shaft is invisible below the surface of the pre-pubic skin and/or scrotum. Such anatomy gives the impression of a short phallus, even though corporeal length is normal and palpable under the overlying skin and subcutaneous tissue.³⁶ In adults, buried penis can result from aging, obesity, complications from an overly aggressive circumcision and, in rare cases, a missed congenital buried penis.^{37,38} In the case of the patient presented, it appears that his buried penis was a combination of aging and obesity.³⁸ The patient had no complications throughout puberty and in early adulthood related to the circumcision; in the last two decades, however, he had difficulty seeing his penis.

The psychological consequences related to a buried penis are well-documented and include distortion of body image, locker room syndrome and functional hindrance of sexual activity perhaps leading to decreased sex drive. Physical consequences related to this condition include painful erection, difficulty with voiding, chronic balanitis and difficulty in maintaining penile hygiene.^{37,39} Although there is no direct link between a buried penis and penile cancer, chronic balanitis and difficulty in maintaining penile hygiene (two consequences of a buried penis) are also identified as risk factors for penile cancer.⁹⁻¹¹

Conclusion

Our case suggests that the development of a buried penis later in life, despite the neonatal circumcision, may pose

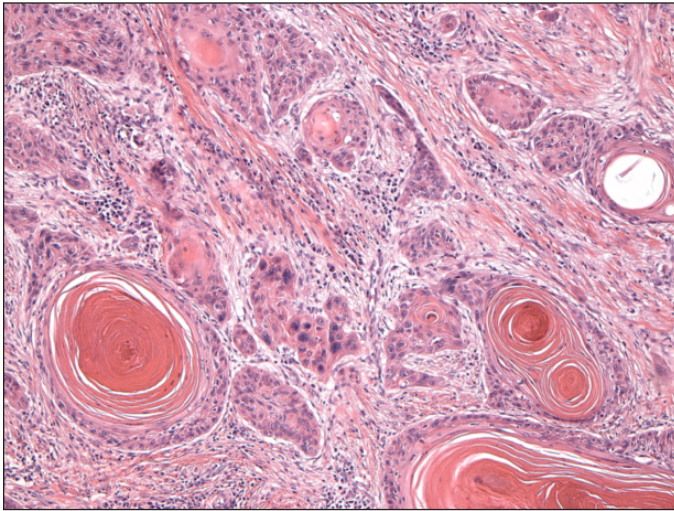


Fig. 2 Infiltrating squamous cell carcinoma showing irregular pattern of malignant squamous cells, focally showing keratin pearls.

a risk factor for the development of penile cancer through such mechanisms as balanitis and poor penile hygiene, both of which are thought to be a sequelae of buried penis and are risk factors for squamous cell cancer of the penis.²⁸⁻³⁰ With the current global obesity epidemic,^{40,41} there will undoubtedly be more patients who present with a buried penis as a result of increasing abdominal girth. Whether they are circumcised or not, proper inspection of the penis in these patients by primary care physicians and specialists will become increasingly important.

Competing interests: None declared.

This paper has been peer-reviewed.

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