

Simultaneous Occurrence of Balanoposthitis Circumscripta Plasmacellularis Zoon, Phimosis and *in Situ* Carcinoma of the Penis: Case Report with An Unusual Ulcerated Polypoid Variant of Zoon's Disease and a Carcinoma *in Situ* of Reserve Cell Type

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Abstract

Citation: Wollina U, Schönlebe J, Goldman A, Tchernev G, Lotti T. Simultaneous Occurrence of Balanoposthitis Circumscripta Plasmacellularis Zoon, Phimosis and *in Situ* Carcinoma of the Penis: Case Report with An Unusual Ulcerated Polypoid Variant of Zoon's Disease and a Carcinoma *in Situ* of Reserve Cell Type. Open Access Maced J Med Sci. <https://doi.org/10.3889/oamjms.2018.020>

Keywords: Balanitis plasmacellularis Zoon; Morbus Queyrat; Penile cancer; Carcinoma *in situ*; Circumcision; Phimosis; HPV

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Received: 28-Jul-2017; **Revised:** 17-Oct-2017;
Accepted: 29-Oct-2017; **Online first:** 31-Dec-2017

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Funding: This research did not receive any financial support

Competing Interests: The authors have declared that no competing interests exist

BACKGROUND: Zoon's balanitis is a benign disease characterized by an asymptomatic, chronic, solitary, shiny, red-orange plaque of the glans and/ or prepuce. In rare cases of Zoon's disease, penile squamous cell carcinoma developed in the chronic inflammatory lesions.

CASE REPORT: We report on a 68-year-old male patient presenting with phimosis and coexistent Zoon's disease and penile carcinoma *in situ* treated successfully by circumcision.

CONCLUSION: Coexistence of both lesions in contrast to the development of cancerous lesions within pre-existent Zoon's disease is a very rare observation.

Introduction

Zoon's balanitis is characterized by an asymptomatic, chronic, solitary, shiny, red-orange plaque of the glans and/ or prepuce. In contrast to Morbus Queyrat which is a carcinoma *in situ*, Zoon's disease is a benign lesion [1]. In rare cases of Zoon's disease, penile squamous cell carcinoma developed in the chronic inflammatory lesions [2][3][4].

The disorder develops in uncircumcised adult to elderly men. Nevertheless, in rare cases, females and circumcised men can be affected [5][6]. Etiology and pathogenesis are not well understood but irritant

contact balanitis is widely accepted [7].

Histologically, the early lesions show a thickened parakeratotic epithelium. Epidermal oedema accompanied by a dense upper dermal band of lympho-histiocytic inflammatory cells including many plasma cells, dilated capillaries, extravasated red blood cells, and hemosiderin deposition develop. There are a greater proportion of IgG4-positive plasma cells in the lesions, but no signs of cicatrication are found. Later on, a thinned and scant spongiotic epithelium occurs, siderophages may be found in the dermis. Subdermal clefts and lozenge keratinocytes can occur. The lesions don't show cytological atypia or epithelial dysplasia [8][9][10]. As

far as we know, Zoon's balanitis is not caused by infection with human papilloma virus (HPV) [11].

Diagnosis is based on clinical pattern and confirmation by histopathology. Noninvasive techniques such as dermoscopy or reflectance confocal microscopy seem to have a potential to differentiate the being lesion from precancerous and cancerous penile imitators [12][13].

Case report

A 68-year-old male patient presented with an asymptomatic reddish papular lesion of the foreskin (Fig. 1). Reposition of the foreskin was not completely possible indicating phimosis.



Figure 1: Strawberry-like reddish papules of the penis – clinical presentation of carcinoma in situ

His medical history was positive for diabetes mellitus and prostate adenoma. Surgical excision in combination with circumcision to correct phimosis was performed with penile root anaesthesia using 1% prilocaine solution (Fig. 2).

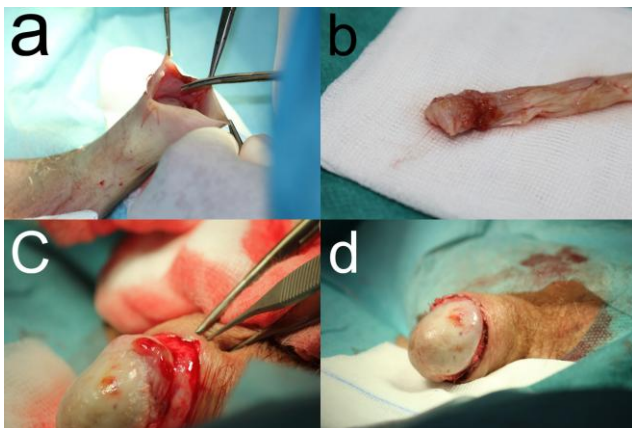


Figure 2: (a) Circumcision of the penis to remove the in situ carcinoma and to correct phimosis; (b) Surgical specimen; (c) Erosive Zoon's disease; (d) After suturing

After removal of foreskin, two shiny reddish ulcerated lesions of the glans penis became visible and were also completely excised (Fig. 2c). The wound was closed with 4/0 absorbable polyglactin sutures (Vicryl rapid®; Ethicon; Norderstedt, Germany) (Fig. 2d). Healing was unremarkable.

Histology: An epidermal in situ carcinoma of the reserve cell type with circumscribed plump taps but complete basal cell membrane (Periodic acid Schiff's reaction and collagen type IV) was observed, associated with a variable dense lichenoid inflammatory infiltrate of the upper dermis (Fig. 3a, b). Locally, hemosiderin depots were seen. R0-resection.

The erosive lesions of the glans penis were characterized as chronic erosive balanoposthitis with a band-like, partly polypoid, and chronic inflammatory reaction, numerous capillaries, surrounded by giant cells. The inflammatory infiltrate was dominated by plasma cells. No epithelial dysplasia, no cytological atypia were observed (Fig. 3 c, d).

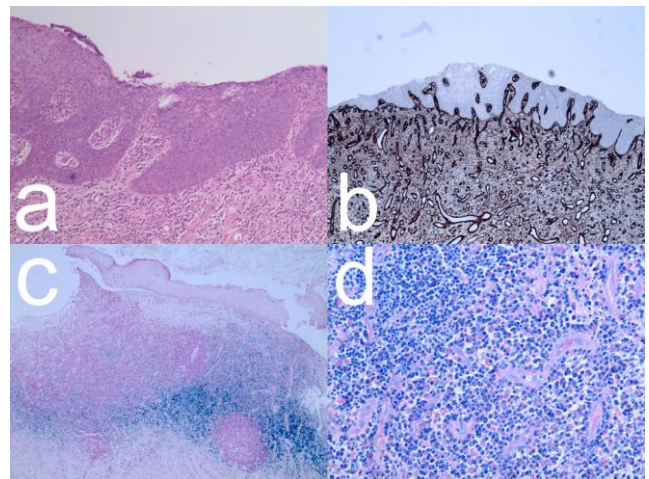


Figure 3: Histopathology. (a) Carcinoma in situ with plump epithelial taps (hematoxylin-eosin x 10). (b) Collagen IV immunoperoxidase staining showing an intact basal cell membrane (x 4). Erosive Zoon's disease with lichenoid dermal inflammatory infiltrate (c; x 4), composed of plasma cells and lymphocytes (d; x 20)

The findings confirmed the diagnoses of penile in situ carcinoma associated with secondary phimosis and ulcerous Zoon's disease.

Discussion

Carcinoma in situ (CIS) of the penis is an uncommon condition among Caucasians, most frequently presenting as red macules or plaques. Early recognition and treatment are important, as progression to invasive penile cancer has been reported in up to 1/3 of cases [14]. European Association of Urology (EAU) guidelines recommend

local excision with or without circumcision, laser therapy with carbon dioxide laser or neodymium:yttrium-aluminium-garnet (Nd: YAG) laser, photodynamic therapy, and topical therapy with 5-FU or 5% imiquimod cream [16].

We performed surgery with circumcision to achieve an R0-status of the cancerous lesion and to correct phimosis in one setting. After circumcision, two ulcerated polypoid lesions, diagnosed as Zoon's disease became visible. We removed them surgically to obtain histologic confirmation. Our differential diagnosis was penile cancer. In case of uncomplicated Zoon's disease, often topical treatment is used primarily.

In contrast to other inflammatory penile disorders, Zoon's disease is usually refractory to topical therapy and systemic antibiotics/ antimycotics. Recently, photodynamic therapy has been used in selected cases but this is not an established treatment [17].

Zoon's disease can be treated relatively easily by circumcision or alternatively by ablative erbium-YAG-laser therapy [18][19]. The latter is a less invasive procedure with no down-time.

The simultaneous occurrence of carcinoma in situ of the reserve cell type and polypoid, ulcerated Zoon's disease hidden by phimosis demonstrates exemplary the diagnostic and therapeutic problems of penile diseases in elderly males. Early diagnosis is of particular importance to avoid invasive penile cancer with severe consequences [20].

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