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

Uwe Wollina1, Jacqueline Schönlebe2, Alberto Goldman3, Georgi Tchernev4,5, Torello Lotti6

1Städtisches Klinikum Dresden - Department of Dermatology and Venerology, Dresden, Sachsen, Germany; 2Städtisches Klinikum Dresden - Institute of Pathology “Georg Schmoro”, Dresden, Germany; 3Clinica Goldman, Plastic Surgery, Porto Alegre/RS, Brazil; 4Medical Institute of Ministry of Interior (MVR), Department of Dermatology, Venereology and Dermatologic Surgery, Sofia, Bulgaria; 5Onkoderma - Polyclinic for Dermatology and Dermatologic Surgery, Sofia, Bulgaria; 6Università Degli Studi “G. Marconi”, Rome, Italy

Abstract

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 cicatrization 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.

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).

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).

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 polyloid 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].

References

1. Zoon J. Balanoposthite chronique circonscrite bénigne à plasmocytes (contra érythroplasie de Queyrat). Dermatológica. 1952;10:51–57.
2. Joshi UY. Carcinoma of the penis preceded by Zoon’s balanitis. Int J STD AIDS. 1999;10(12):823-5. https://doi.org/10.1258/095646299193484 PMId:10639067
3. Porter WM, Hawkins DA, Dinneen M, Bunker CB. Zoon’s balanitis and carcinoma of the penis. Int J STD AIDS. 2000;11(7):484-5. PMId:10191496
4. Balato N, Scalvenzi M, La Bella S, Di Costanzo L. Zoon’s balanitis: benign or premalignant lesion? Case Rep Dermatol. 2009;1(1):7-10. https://doi.org/10.1159/000210440 PMId:20652106 PMcId:PMC2895202
5. Adégbidi H, Atadokpédé F, Dégbocé B, Saka B, Akpadjan F, Yadomon H, Padonou Fdo A. [Zoon’s balanitis in circumcised and HIV infected man, at Cotonou (Benin)]. Bull Soc Pathol Exot. 2014;107(3):139-41. https://doi.org/10.1007/s13149-014-0359-4 PMId:24792459
6. Yoganathan S, Bohl TG, Mason G. Plasma cell balanitis and vulvitis (of Zoon). A study of 10 cases. J Reprod Med. 1994;39(12):939-44. PMId:7884748
7. Dayal S, Sahu P. Zoon balanitis: A comprehensive review. Indian J Sex Transm Dis. 2016;37(2):129-138. https://doi.org/10.4103/0925-7184.192128 PMId:27890945 PMcId:PMC5112996
8. Alessi E, Coggi A, Gianotti R. Review of 120 biopsies performed on the balanopreputial sac. from zoon’s balanitis to the concept of a wider spectrum of inflammatory non-cicatricial balanoposthitis. Dermatology. 2004;208(2):120-4. https://doi.org/10.1159/000076848 PMId:15057000
9. Kumar B, Naranj T, Dass Radotra B, Gupta S. Plasma cell balanitis: clinicopathologic study of 112 cases and treatment modalities. J Cutan Med Surg. 2006;10(1):10-9. https://doi.org/10.1016/j.jcms.2006.0008 PMId:17241566
10. Aggarwal N, Panwani AV, Ho J, Cook JR, Sverdlow SH. Plasma cell (Zoon) balanitis: another inflammatory disorder that can be rich in IgG4+ plasma cells. Am J Surg Pathol. 2014;38(10):1437-43. https://doi.org/10.1097/PAS.0000000000000269 PMId:25216321
11. Kiene P, Fölster-Holst R. No evidence of human papillomavirus infection in balanitis circumscripta plasmacellularis Zoon. Acta Derm Venereol. 1995;75(6):496-7. PMId:8651036
12. Arzberger E, Komerici P, Ahlgirm-Siess V, Massone C, Chubisov D, Hofmann-Wellenhof R. Differentiation between balanitis and carcinoma in situ using reflectance confocal microscopy. JAMA Dermatol. 2013;149(4):440-5. https://doi.org/10.1001/jamadermatol.2013.2440 PMId:2325422
13. Corazza M, Virgili A, Minghetti S, Toni G, Borghi A. Dermoscopy in plasma cell balanitis: its usefulness in diagnosis and follow-up. J Eur Acad Dermatol Venereol. 2016;30(1):182-4. https://doi.org/10.1111/jdv.12692 PMId:25176141
14. Mikhail GR. Cancers, precancers, and pseudocancers in the male genitalia. A review of clinical appearances, histopathology, and management. J Dermatol Surg Oncol. 1996;6(12):1027–35. https://doi.org/10.1177/0146946896060102
15. Hakenberg OW, Compérat EM, Minhas S, Necchi A, Protzel C, Wollina U. Ablative erbium: YAG laser treatment of idiopathic cicatricial balanoposthitis (Zoon’s). Acta Derm Venereol. 1995;75(6):496-7. PMId:8651036
16. Torchia D, Cappugi P. Photodynamic therapy for Zoon balanitis. Eur J Dermatol. 2014;24(6):707. PMId:25333228
17. Edwards SK, Bunker CB, Ziller F, van der Meijden WJ. 2013 European guideline for the management of balanoposthitis. Int J STD AIDS. 2014;25(9):815-26. https://doi.org/10.1177/0956462414533099 PMId:24828553
18. Wollina U. Erbium-YAG-laser therapy of balanitis circumscripta plasmacellularis Zoon. Med Laser Appl. 2006;21(1):23-6. https://doi.org/10.1016/j.ml.a.2005.11.002
19. Wollina U. Ablative erbium: YAG laser treatment of idiopathic chronic inflammatory non-cicatricial balanoposthitis (Zoon’s disease) – A series of 20 patients with long-term outcome. J Cosmet Laser Ther. 2010;12(3):120–3. https://doi.org/10.3109/14764171003706125 PMId:20429688
20. Wollina U, Steinbach F, Verma S, Tchernev G. Penile tumours: a review. J Eur Acad Dermatol Venereol. 2014;28(10):1267-76. https://doi.org/10.1111/jdv.12491 PMId:24684263