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Zoon's Balanitis (Balanitis Circumscripta Plasmacellularis) – a Case Report with a Review of Literature

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Abstract: Zoon's balanitis or balanitis circumscripta plasmacellularis is a rare, idiopathic, chronic, benign, inflammatory disease of the genital mucosa of non-sexually transmitted nature. Etiopathogenesis is not fully understood, but the moist environment and chronic irritation are considered as important triggers. It occurs mainly in uncircumcised middle- and elderly men, but there are single cases in circumcised men. Affects the glans penis and foreskin. It is also rarely seen in women, such as Zoon's vulvitis, with predilectation involvement of labia minora. The article presented an overview of epidemiology, clinical performance, histopathological characteristics, diagnostic criteria and diagnostic methods of Zoon's balanitis. In addition, the differential diagnosis of the disease is discussed. Treatment methods are considered and circumcision is highlighted as the "gold standard" and first choice. We present a 53-year-old man with a well-circumscribed erythematous plaques on the glans penis and the inner surface of the foreskin from 10 years. Histologically plasmocyte infiltrate is established. Based on the correlation of history, dermatological status and histopathological result was diagnosed Zoon's balanitis. Conservative topical therapy was our preferred way of treatment. On an outpatient basis, we recommended personal genital hygiene, mometasone furoate cream 0.1% for 2 weeks and tacrolimus 0.1% ointment for 1 month. A control examination after 1 and a half months found a good clinical result with mild residual erythema. The patient was explained both the need for regular clinical and histological monitoring due to the carcinogenic potential of the lesions and the option of therapeutic circumcision.

Keywords: Zoon Balanitis, Circumcision, Nonvenereal Disease, Topical Calcineurin Inhibitors, Lasers

1. Introduction

Zoon's balanitis (ZB) or balanitis circumscripta plasmacellularis is a benign, non-sexually-transmitted inflammatory disease of the genital mucosa [1]. It is considered an idiopathic condition that often occurs in non-circumcised men [1]. Usually it follows a chronic persistent course [2]. Clinically, a solitary, well-circumscribed erythematous plaque with a shiny surface, which is most commonly on a glossy penis is presented [1, 2]. In rare cases, it may also occur in women (Zoon's vulvitis or plasma cell vulvitis) [3].

2. Clinical Case

A 53-year-old man with redness and periodic secretion in the glans area of the penis and foreskin with a duration of ten years is presented. Some edema and "sores" appeared in the last 2 months. The lesions were moderately painful. He

underwent long-term outpatient topical anti-mycotic and antibiotic treatment with no effect. His medical history revealed arterial hypertension, compensated with ramipril 5 mg/day and bisoprolol 5 mg/day. Slightly elevated fasting glucose levels were found on routine blood tests, for which dietary restrictions were recommended. The dermatological examination showed well-demarcated erythematous plaques with glossy surface, varying 1.5–2 cm in diameter, surrounded by peripheral oedema with single vesicules on the glans penis and the inner surface of the foreskin (Figure 1). The histopathological examination revealed acanthosis and balloon degeneration of the epithelium, extensive papillary oedema and abundant perivascular infiltrate of plasma cells in tunica propria (Figure 2, Figure 3). Based on the clinico-pathological correlation, Zoon's balanitis was diagnosed. Topical treatment with mometasone furoate cream 0.1% for 2 weeks, followed by tacrolimus 0.1% ointment for 1 month was introduced with a good clinical outcome. The possibility of conducting circumcision or laser treatment (Carbon dioxide laser (CO2)/Erbium: YAG laser) was discussed together with the need of close clinical and histopathological follow-up.

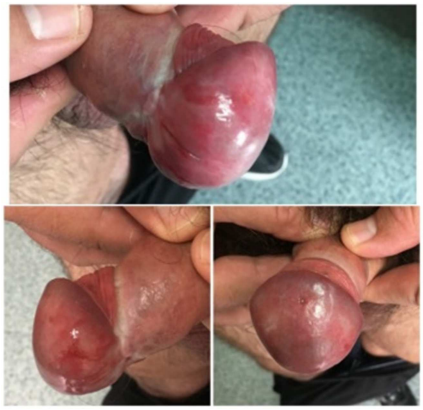

Figure 1. *Well-demarcated erythematous plaques with shiny surface and dimensions up to 1.5 – 2 cm, circumscribed by oedema and tiny vesicles on the gloss penis and the inner surface of the foreskin.*

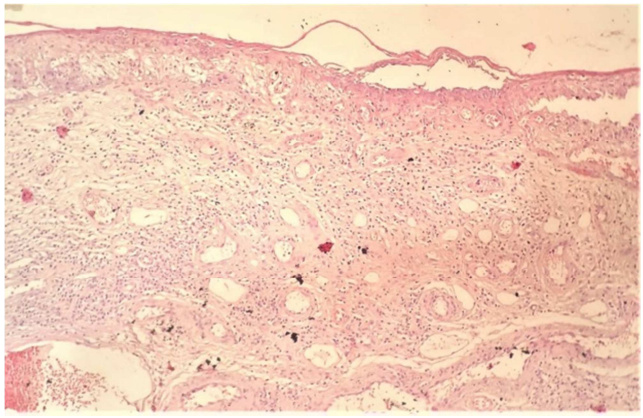

Figure 2. *HE staining (x100): acanthosis, balloon degeneration of the lower epithelium with massive plasma cell exocytosis; diffuse interstitial and perivascular infiltration in tunica propria.*

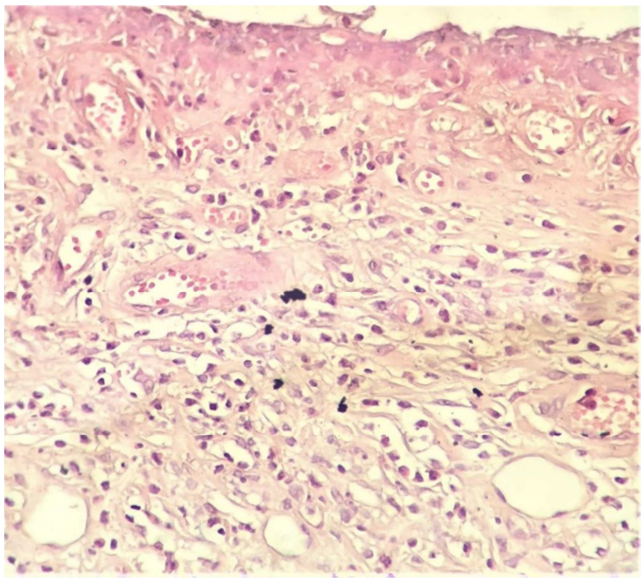

Figure 3. *HE staining (x 200): diffuse plasma cell-rich inflammatory infiltrate in tunica propria.*

3. Discussion

ZB was first described by the Dutch dermatologist J. J. Zoon in 1952 [4]. It is a rare condition, affecting mostly uncircumcised middle- and elderly men. The age range is 20 to 91 years [5, 6]. Anecdotal cases are reported in circumcised men, too [7, 8]. The etiology is not fully understood [1, 5, 6]. Factors of primary importance are humid environment and chronic irritation [6]. Urinary retention and smegma, in the context of the so-called "dysfunctional foreskin", is associated with poor genital hygiene and recurrent infections, as in our patient [1, 9]. Possible trigger factors are trauma, friction and heat [5, 9]. Some authors considered chronic infection with *Mycobacterium smegmatis* and *Human papillomaviruses* (HPV) as an etiological cause [1].

In the greatest percentage, lesions are localized on the gloss penis and the inner surface of the foreskin [10]. The oral mucosa, conjunctiva, epiglottis, urethra and cheeks are rarely affected [11]. In females they are located on labia minora [3, 12]. The erythematous, well-circumscribed lesions usually proceed the correct verification for more than 2 years [10, 13]. They are often asymptomatic. Sometimes, accompanying itching, dysuria, increased sensitivity, burning and pain may evolve [1]. Bleeding or dyspareunia (painful or difficult intercourse) has been reported rarely [14].

The disease presents with circumscribed erythematous patches with shiny and moist surface, which can be single or multiple [2, 8, 10]. Multiple point-like, bright red spots - "cayenne pepper spots" - are sometimes observed on the surface that result from microhemorrhages and hemosiderin deposition [1, 15]. In the foreskin area, so-called "kissing lesion", may arise from direct contact with closely spaced lesions [1, 15]. When eroding surface is described as the "rusty stain" ("rusty spot") phenomenon [1, 15]. ZB can also occur in the form of a vegetative or "multiple" clinical variant [6].

The diagnostic milestones are suggested by B. Kumar et al. [10] and include: shiny erythematous plaque in the gloss penis area, foreskin, or both; persistence of lesions for more than three months; absence of lesions, characteristic of lichen planus or psoriasis in the predilection sites of the skin; poor response to topical treatment (minimum 4 weeks); absence of concomitant infections excluded by additional laboratory tests: Gram staining, Tzanck test, potassium-based sample and venereological tests. A minimum of three of the five criteria are required for diagnosis [1].

Histopathological changes affect both the epidermis and dermis [1]. Acanthosis and parakeratosis are found, followed by atrophy, erosions and spongiosis [1, 6, 11]. In the early stages, lichenoid infiltrate from lymphocytes and single plasmocytes are observed in the papillary dermis. Subsequently, dense infiltrate of plasmocytes, neutrophils, eosinophils, lymphocytes and erythrocytes is found. Normally, plasmocytes predominate in more than 50% of the inflammatory cell population [1, 6, 11]. Changes in dermal vessels are represented by vascular dilatation with vertical or oblique orientation of individual proliferated vessels, which is characteristic of ZB. In cases of "cayenne pepper spots" erythrocyte extravasation and hemosiderin deposition are present [6]. The diagnostic histopatological criteria include four main parameters: (1) epidermal edema; (2) dense infiltrate from chronic inflammatory cells, including many plasma cells in the papillary dermis; (3) dilated capillaries and extravased erythrocytes; (4) deposition of hemosiderin [10]. Immunohistochemically, plasma cells in ZB were found to produce predominantly immunoglobulins of class IgG and less IgA and IgM [1, 2].

Some authors have found an association between ZB and Queyrat erythroplasia, while others suggest that ZB may precede penile carcinoma, since carcinogenic changes may occur in the chronically inflamed mucosa of ZB. Four cases have been published, three with Queyrat's erythroplasia and one with penile carcinoma, occurring on ZB lesions [16-19]. In addition, a case of lichen sclerosus has been reported to develop in a patient with ZB after treatment with a carbon dioxide laser [2]. Multicenter longitudinal studies with more patients are needed to prove these associations.

ZB differential diagnosis is extremely extended [1]. Consideration includes dermatoses with mucosal involvement, mycoses, dermatovirosis, aerobic, anaerobic and mycobacterial infections [1, 13, 14]. Individual forms of balanitis should also be discussed, as should Reiter's syndrome [14]. Due to the fact that ZB is regarded as a precancerous state, the most important is the distinction from Erythroplasia of Queyrat [squamous cell carcinoma in situ (CIS)], penile carcinoma and Bowen's disease [1, 14, 16-18]. Histopathologically, dysplastic epithelium is found in the precancerous lesions [1]. Reflected confocal microscopy (RCM)] helps to differentiate ZB from CIS [19]. The characteristic findings in CIS are atypical cell arrangement in the form of honeycomb, disorganized dermo-epidermal margin and round cells with large nuclei [19]. Standard dermatoscopy is also cited as a reliable method to differentiate ZB from premalignant/malignant conditions [20]. In ZB, structureless foci or diffuse yellow-orange areas are found as a result of hemosiderin deposition, as well as curved blood vessels corresponding to vascular dilation and proliferation [21-23]. Less commonly, linear irregular blurred vessels and sperm-like vessels are found [24-27]. In Queyrat erythroplasia, there are scattered glomerular vessels, in psoriasis - regular dotted glomerular vessels, and in non-specific balanitis often linear, irregular, non-specific, blurry, unfocused vessels are seen [28-30].

ZB treatment is complex [14]. Good daily personal hygiene is mentioned as a measure of primary importance [14]. The topical therapy includes mild to moderate corticosteroid, calcineurin inhibitors, antibiotic preparations and immune-modulators such as imiquimod. Carbon dioxide or erbium: YAG laser coagulation is considered in therapeutically refractory patients [31-33]. Photodynamic treatment is also beneficial [34, 35]. Gentian violet, antimycotics, cryotherapy [36], electrodesication, with no or minor effect were applied topically. An effect on vulvar lesions by intralesional interferon-α was found [37]. Some cases have responded to systemic therapy with griseofulvin, fusidic acid, dapsone, or corticosteroids [1, 36]. However, circumcision is the "gold standard" therapeutic option according to the 2013 European guidelines [14].

4. Conclusion

ZB is rare venereal condition with obscure etiology and pathogenesis. It present an enormous diagnostic challenge due to non-specific clinical picture, recalcitrant course and unclear biological potential with possible malignant degeneration. A great alert exists in all patients with long-lasting erythematous plaque with glossy surface on the penile area, refractory to topical treatment. A close clinico-pathological follow-up is always recommended in all such cases.

References

[1] Dayal S, Sahu P. Zoon balanitis: A comprehensive review. Indian J Sex Transm Dis AIDS. 2016; 37 (2): 129-138.

[2] Retamar RA, Kien MC, Chouela EN. Zoon's balanitis: Presentation of 15 patients, five treated with a carbon dioxide laser. Int J Dermatol. 2003; 42: 305-307.

[3] Damiani L, Quadros M, Posser V, et al. Zoon vulvitis. An Bras Dermatol. 2017; 92 (5 Suppl 1): 166-168.

[4] Zoon JJ. Chronic benign circumscript plasmocytic balanoposthitis. Dermatologica. 1952; 105: 1–7.

[5] Balato N, Scalvenzi M, La Bella S, et al. Zoon's Balanitis: Benign or Premalignant Lesion? Case Rep Dermatol. 2009; 1 (1): 7-10.

[6] Lepe K, Salazar FJ. Balanitis Circumscripta Plasmacellularis (Plasma Cell Balanitis, Zoon Balanitis) [Updated 2019]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2020. Available from: https://www.ncbi.nlm.nih.gov/books/NBK482129/

[7] Toker SC, Baskan EB, Tunali S, et al. Zoon's balanitis in a circumcised man. J Am Acad Dermatol. 2007; 57 (Suppl 2): 6-7.

[8] Adégbidi H, Atadokpèdé F, Dégboé B, et al. [Zoon's balanitis in circumcised and HIV infected man, at Cotonou (Benin)]. Bull Soc Pathol Exot. 2014; 107 (3): 139-141.

[9] Porter WM, Bunker CB. The dysfunctional foreskin. Int J STD AIDS. 2001; 12: 216-220.

[10] Kumar B, Narang T, Dass Radotra B, et al. Plasma cell balanitis: Clinicopathologic study of 112 cases and treatment modalities. J Cutan Med Surg. 2006; 10: 11-15.

[11] Weyers W, Ende Y, Schalla W, et al. Balanitis of Zoon: A clinicopathologic study of 45 cases. Am J Dermatopathol. 2002; 24: 459-467.

[12] Jaimes Suarez J, Vidal Conde L, Collazos Robles R, et al. Zoon Vulvitis Treated Successfully With Platelet-Rich Plasma: First Case Reported. J Low Genit Tract Dis. 2017; 21 (4): 48-51.

[13] Altmeyer P, Kastner U, Luther H. Balanitis/balanoposthitis chronic circumscripta benigna plasmacellularis - entity or fiction?. Hautarzt. 1998; 49 (7): 552-555.

[14] Edwards SK, Bunker CB, Ziller F, et al. 2013 European guideline for the management of balanoposthitis. Int J STD AIDS. 2014; 25: 615-626.

[15] Pastar Z, Rados J, Lipozencic J, et al. Zoon plasma cell balanitis: An overview and role of histopathology. Acta Dermatovenerol Croat. 2004; 12: 268-273.

[16] Starritt E, Lee S. Erythroplasia of Queyrat of the glans penis on a background of Zoon's plasma cell balanitis. Australas J Dermatol. 2008; 49: 103-105.

[17] Joshi UY. Carcinoma of the penis preceded by Zoon's balanitis. Int J STD AIDS. 1999; 10: 823-825.

[18] Divakaruni AK, Rao AV, Mahabir B. Erythroplasia of Queyrat with Zoon's balanitis: A diagnostic dilemma. Int J STD AIDS. 2008; 19: 861-863.

[19] Arzberger E, Komericki P, Ahlgrimm-Siess V, et al. Differentiation between balanitis and carcinoma in situ using reflectance confocal microscopy. JAMA Dermatol. 2013; 149: 440-445.

[20] Errichetti E, Lacarrubba F, Micali G, et al. Dermoscopy of Zoon's plasma cell balanitis. J Eur Acad Dermatol Venereol. 2015; 30: 2137-2139.

[21] Yoganathan S, Bohl TG, Mason G. Plasma cell balanitis and vulvitis (of Zoon). A study of 10 cases. J Reprod Med. 1994; 39: 939-944.

[22] Tang A, David N, Horton LW. Plasma cell balanitis of Zoon: Response to Trimovate cream. Int J STD AIDS. 2001; 12: 75-78.

[23] Santos-Juanes J, Sánchez del Río J, Galache C, et al. Topical tacrolimus: An effective therapy for Zoon balanitis. Arch Dermatol. 2004; 140: 1538-1539.

[24] Hernandez-Machin B, Hernando LB, Marrero OB, et al. Plasma cell balanitis of Zoon treated successfully with topical tacrolimus. Clin Exp Dermatol. 2005; 30: 588-589.

[25] Kyriakou A, Patsatsi A, Patsialas C, et al. Therapeutic efficacy of topical calcineurin inhibitors in plasma cell balanitis: Case series and review of the literature. Dermatology. 2014; 228: 18-23.

[26] Bardazzi F, Antonucci A, Savoia F, et al. Two cases of Zoon's balanitis treated with pimecrolimus 1% cream. Int J Dermatol. 2008; 47: 198-201.

[27] Stinco G, Piccirillo F, Patrone P. Discordant results with pimecrolimus 1% cream in the treatment of plasma cell balanitis. Dermatology. 2009; 218: 155-158.

[28] Petersen CS, Thomsen K. Fusidic acid cream in the treatment of plasma cell balanitis. J Am Acad Dermatol. 1992; 27: 633-634.

[29] Nasca MR, De Pasquale R, Micali G. Treatment of Zoon's balanitis with imiquimod 5% cream. J Drugs Dermatol. 2007; 6: 532-534.

[30] Marconi B, Campanati A, Simonetti O, et al. Zoon's balanitis treated with imiquimod 5% cream. Eur J Dermatol. 2010; 20: 134-135.

[31] Baldwin HE, Geronemus RG. The treatment of Zoon's balanitis with the carbon dioxide laser. J Dermatol Surg Oncol. 1989; 15: 491-494.

[32] Albertini JG, Holck DE, Farley MF. Zoon's balanitis treated with Erbium: YAG laser ablation. Lasers Surg Med. 2002; 30: 123-126.

[33] Wollina U. Erbium-YAG-laser treatment of balanoposthitis chronica circumscripta benigna plasmacellularis Zoon. Med Laser Appl. 2006; 21: 23-26.

[34] Pinto-Almeida T, Vilaça S, Amorim I, et al. Complete resolution of Zoon balanitis with photodynamic therapy – A new therapeutic option? Eur J Dermatol. 2012; 22: 540-541.

[35] Borgia F, Vaccaro M, Foti A, et al. Zoon's balanitis successfully treated with photodynamic therapy: Case report and literature review. Photodiagnosis Photodyn Ther 2016; 13: 347-349.

[36] Bari O, Cohen PR. Successful Management of Zoon's Balanitis with Topical Mupirocin Ointment: A Case Report and Literature Review of Mupirocin-Responsive Balanitis Circumscripta Plasmacelluaris. Dermatol Therapy. 2017; 7: 203–210.

[37] Neri I, Patrizi A, Marzaduri S, et al. Vulvitis plasmacellularis: two new cases. Genitourin Med. 1995; 71 (5): 311-313. doi: 10.1136/sti.71.5.311.