

CASE REPORT

Zoon's Balanitis. A case report

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ABSTRACT

Zoon's balanitis (ZB) is a benign inflammatory mucositis of the head of the penis (glans penis), which generally manifests in uncircumcised middle-aged and elderly men. Zb's course is usually chronic. Sometimes recurrent.

A 21-year-old uncircumcised male patient presents with a 3-year clinical picture characterized by the presence of an erythematous macule and two infiltrated patches on the foreskin's inner face and gland. As a consequence, a biopsy was conducted. It reported an eroded tissue structure with granulation tissue development and lymphoplasmocitary accentuated infiltrate, confirming ZB diagnosis. Treatment with oral and topical corticosteroids, associated to pimecrolimus, was administered. There was almost complete resolution of the lesions.

INTRODUCTION

This entity was first described by Zoon in 1952 in eight patients who presented chronic balanitis with similar histopathologic features. Consequently, it was acknowledged as "balanitis circumscripita plasmacellularis." It consists of an inflammatory, benign, idiopathic mucositis, with a tendency to chronicity due to its capacity to persist for months or years. It usually manifests as a well-defined, erythematous and solitary plaque on the gland of middle-age (or older) non-circumcised males.^{1,2}

Histopathology reveals a plasmatic cell inflammatory infiltrate, epidermal atrophy, spongiosis, tortuous blood vessels and inflammatory cell exocytosis.³

Risk factors include smoking and bad genital hygiene.⁴ Treatments include circumcision, hygiene measures,

topical steroids with or without antibiotics, CO2 laser and topical tacrolimus.⁵

CLINICAL CASE

The case of a 21-year-old uncircumcised male patient is presented. There is no significant personal pathologic history, but rather a 3-year clinical picture characterized by two infiltrate, deep red, wet patches with irregular margins and erosive surface, located on the foreskin's inner face, and a slightly erythematous and poorly defined macular lesion on the gland. (Fig. 1 and 2)

Dermoscopy showed multiple irregular linear vessels, serpentine vessels, convoluted vessels and some spermatozoa-like vessels. (Fig. 3 and 4)

A biopsy and histopathology study were conducted. The reports showed tissue eroded structure with granulation tissue development, lymphoplasmocitary accentuated infiltrate and, to a lesser degree, the presence of neutrophils. There are no neoplastic changes. (Fig. 5)



Figure 1: Irregular infiltrate erythematous plaques with erosive surface, located on inner face of foreskin.



Figure 2: Slightly erythematous and poorly defined macular lesion on the gland.

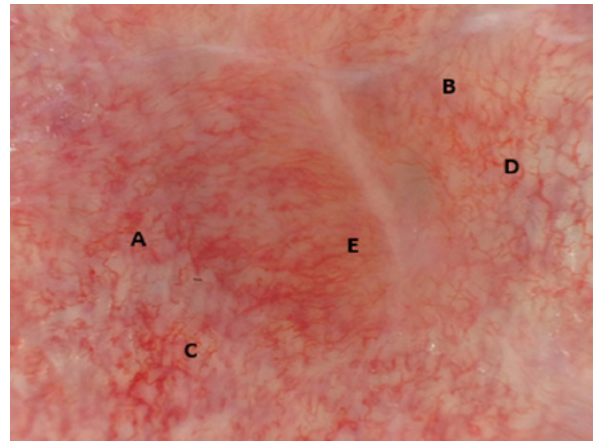


Figure 3: Dermoscopy of gland lesion. A serpiginous vessels, B irregular linear vessels, C convoluted vessels, D spermatozoa-like vessels, E orange areas.

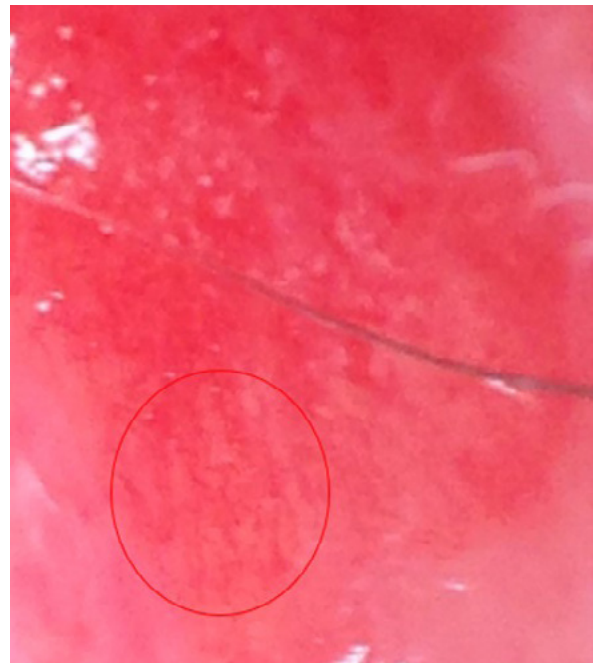


Figure 4: Irregular linear vessels on foreskin lesion.

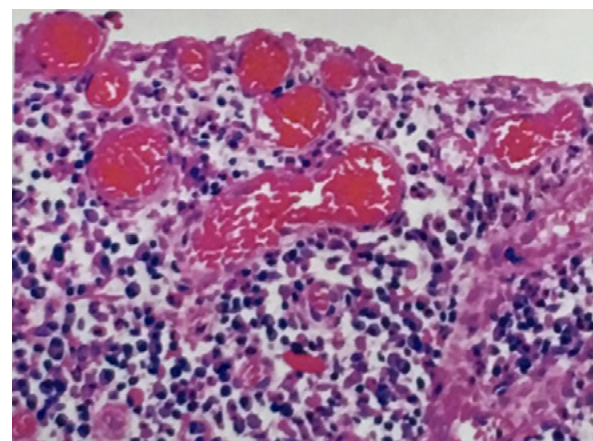


Figure 5: Mainly plasmocitary infiltrate.

Clinical and histopathological findings confirmed BZ diagnosis.

Oral prednisone was administered in a dose of 20 mg a day during five days. Moreover, treatment included topical mometasone, followed by pimecrolimus (15 days each). This resulted in full resolution of lesions after a month, without any relapse.

DISCUSSION

Zoon's balanitis (ZB) is a non-venereal dermatosis which manifests mainly in middle-age men and elderly individuals. There is little to no information about the prevalence and incidence of this entity. Mallon and collaborators reported 27 cases of BZ in 357 men diagnosed with pathology of genital skin. Pearce et al presented 26 cases involving 226 patients with BZ, evaluated in a genitourinary medicine clinic. However, there are missing statistics about the presence of the illness on medical literature.²

Regarding the etiopathogeny, it is believed that factors, such as uncircumcision and poor genital hygiene, are involved. These cause urine and smegma retention which generates recurrent local infection that, along with heat and constant friction, increase the possibilities of the condition's manifestation. Some authors suggest the participation of infectious agents, such as *Mycobacterium smegmatis* and the human papillomavirus (HPV). However, no bacterial infections have been reported yet. Moreover, no human papillomavirus' DNA has been detected in the polymerase chain reaction.^{2,3} There is a report about a circumcised patient. Clinical and histopathologic diagnosis concurs with ZB. Such finding is attributed to the fact that the procedure was deemed as inadequate. Consequently, circumcision plays a big part on the prevention of this disorder.⁶

The clinical presentation corresponds to one or more deep red plaques, which may adopt a raised or macular form, along with a wet appearance on the glans, balanoprepucial groove and inner face of the foreskin, with a tendency to erode, ulcer and bleed; frequently, it manifests multiple red bright dots, known as "cayenne

pepper spots," on the background of such plaque. This is due to microhemorrhage and hemosiderin deposit. These findings are accompanied by dysuria, pruritus and a local burning sensation.^{2,7}

Dermoscopy tends to act as a useful method to improve non-invasive diagnoses for this type of balanitis, from which one can observe signs described as a diffuse, yellowish-orange area without focal structure, caused by the deposit of hemosiderin, serpiginous curved vessels, convoluted vessels, goblet-shaped vessels due to dilatation and vascular proliferation. Vascular lesions, similar to spermatozoa, have also been described, along with punctiform and irregular linear vessels at a minor rate.⁸ The presence of the plasmocitary infiltrate on the histopathology does not visually translate on the dermoscopy.⁹

The patient presented with clinical and dermoscopic findings suggestive of ZB (confirmed by the biopsy). However, because this pathology is not as common in young people, one must acknowledge other causes for genital lesions in order to adequately identify the pathology and treatment.

It is crucial to make a clinical differentiation between ZB and other entities, such as erythroplasia of Queyrat, fixed drug eruption, psoriasis, lichen planus, seborrheic dermatitis, secondary syphilis, candidiasis and Reiter's syndrome.¹

Dermoscopic differential diagnosis includes erythroplasia of Queyrat, which shows disperse glomerular vessels; lichen planus, which presents with Wickham white striae and psoriatic balanitis showing a regular homogenous vascular pattern, formed by small punctiform vessels or a denser vascular pattern with tortuous dilated capillaries. Moreover, it is essential to acknowledge that "spermatozoa-like" vessels are not exclusive of ZB, as they have been discovered in some cases of mycosis fungoides as well.⁹

Histopathology is characterized by epidermal atrophy, rhomboidal keratinocytes, spongiosis, inflammatory cell exocytosis and dense subepidermal infiltrate composed mostly by plasma cells, along with extravasation

of erythrocytes, hemosiderin deposit in some occasions and the presence of tortuous vessels.^{1,3}

In 2006, Kumar and collaborators published some clinical criteria for ZB diagnosis: Bright erythematous plaques on the glans, foreskin or both, lesions that evolve during a period greater than three months, absence of lesions suggestive of lichen planus or psoriasis in any part of the body, bad response to toxic therapy (a minimum of four weeks), absence of infectious diseases, excluded by the Tzanck test, potassium hydroxide, Gram stain and VDRL.^{10,1} The patient complied with four of these criteria due to the evolution period of the disease (three years). He presented with lesions located on the glans and the foreskin's inner face, as well as absence of any genital infectious disease and lesions suggestive of inflammatory disorders in other areas of the body.

Treatment aims to improve genital hygiene conditions, circumcision, which leads to the resolution of lesions, as well as the use of topical preparations based on corticosteroids with or without antibiotics. CO2 laser has been present in these cases.⁵

In his 2018 report, Kumar presents with eighteen non-circumcised male patients with ZB diagnosis, who accepted to be circumcised and were monitored for 5 years with no sign of any relapse. Circumcision is considered as a first-line treatment because it eliminates the occlusion produced by the foreskin and inflammation caused by smegma and chronic friction.¹¹

In regards to the use of calcineurin inhibitors, there is certain concern for the relation of these and carcinogenesis. Consequently, it is recommended to rule out any malignant or premalignant lesion before starting the treatment to avoid neoplastic invasion and evolution.¹² Bardazzi's report about two patients treated with pimecrolimus indicates that one of them had full resolution of lesions and the other patient greatly improved the lesions, leaving only a residual hyperpigmented patch.

There are more reports on cases involving treatment with topical pimecrolimus for periods of four to six weeks,

resulting in good tolerance levels and remission of the clinical picture, labeling it as a therapeutic option for patients who do not wish to be surgically intervened.^{14,15}

Another study of 10 patients with ZB showed favorable results regarding topical treatment in combination with 3% oxytetracycline, nystatin and 0.05% clobetasone butyrate, which may suggest topical corticosteroids play an important role on the treatment of this particular condition.

Among other therapeutic options are 5% imiquimod, fusidic acid, dapsone, photodynamic therapy and non-ablative Erbium-Yag laser.¹⁰

CD20 expression of immunohistochemistry is a finding which opens up possibilities for a new type of alternative treatment, such as Rituximab.¹⁷

There are few case reports within the medical literature about young patients and children with ZB.¹⁸ De Oliveira et al. present the case of an adolescent with vertical transmission HIV, who was circumcised. Notable improvements were observed.¹⁹

Lastly, the association between ZB and malignancy is rare. Nonetheless, if existent, it is due to a reactive process caused by subyacent neoplasia. Follow-up on these patients is important in order to detect this potential phenomena at an early stage.²⁰

CONCLUSION

The importance of this case lies on how unusual this presentation of BZ is on young patients; so much that there are little to no published literature about it. Moreover, the excellent response obtained from combined topical treatment must be acknowledged too.

Uncircumcision as a predisposing factor, and clinical and histopathological findings led physicians to the diagnosis of BZ. Within treatment, circumcision seems to be the first-line treatment. However, other alternatives must be available in case patients do not agree.

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