Balanitis Circumscripta Plasmacellularis

—An Unusual Presentation—

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A 79-year-old man with balanitis circumscripta plasmacellularis (BCP), presenting as an erythematous constricting band of the inner surface of the prepuce encircling the penile shaft is described. The biopsy specimen of the lesion showed, in addition to the typical histologic findings of BCP, increased fibrosis and decreased amount of elastic fibers which correlate well with our clinical observations. Electron microscopic examination revealed no viral particles or elastic fibers. Immunohistologically, IgG was found to be the major immunoglobulin class in the plasma cellular infiltrate. (Ann Dermatol 3: (2) 158–163, 1991)

Key Words : Balanitis circumscripta plasmacellularis, Constricting band

Balanitis circumscripta plasmacellularis (BCP) was first described by Zoon1 in 1952 as benign penile erythroplasia without any evidence of malignant potential, thus differentiating it from erythroplasia of Queyrat. It is characterized, clinically, by brick-red, glistening, macular erythema and erosion with multiple, pin-point, bright red spots on the glans and prepuce; histologically, by a dense plasmacytic infiltrate in the dermis.

Although there may be a variety of diseases involving true mucous membranes and mucocutaneous junctions which on biopsy show a dense, subepidermal, plasmacytic infiltrate, it has been stated that BCP is a unique clinical entity, occurring preferentially in elderly uncircumcised men, with a chronic course, unaffected by conventional treatment.

We report, herein, an interesting case of an elderly man with BCP, who presented with an erythematous, erosive, constriction band of the inner surface of the prepuce encircling the penile shaft. To our knowledge, this type of cutaneous lesion has not been previously reported in patients with BCP.

REPORT OF A CASE

A 79-year-old man was first seen at our outpatient clinic in July, 1989. His chief complaint was a 6-month history of painful erosion on the prepuce. He was using no topical or systemic medications during the preceding months, specifically denying any previous trauma history. Initial examination revealed an ovoid 3- to 6- mm shallow erosion with surrounding erythema around the inner surface of the prepuce(Fig. 1). He was uncircumcised and the regional lymph nodes were not enlarged. Cultures and

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special stains for fungi and bacteria were negative. Clinically, a genital sore and erythrolaspsia of Queyrat were suspected and a skin biopsy was performed on the erosive lesion of the prepuce. The biopsy specimen was read as "nonspecific". Initial treatment with topical hydrocortisone cream and oral antibiotics proved unrewarding.

In September 1990, he was seen again with an exacerbation of the previous penile skin lesion. On examination, an area of shiny, glazed, macular erythema with multiple pin-point bright red spots was seen on the glans penis (Fig. 2). Interestingly, the erythematous erosive lesion of the inner surface of the prepuce progressed to form a constricting band encircling the penile shaft (Fig. 3). On erection, this lesion gave him a squeezing pain sensation. Inguinal and generalized lymphadenopathy were absent. A potassium hydroxide preparation gave negative results for fungal elements.

The results of the following laboratory examination were negative or within normal limits: complete blood count with differential count, urinalysis, VDRL, TPHA and liver function test. But ESR was elevated at 42 mm/hr. The second biopsy was performed on the erythematous constricting band of the inner surface of the prepuce. Microscopic examination revealed a thin epidermis with a focal absence of horny and granular layer. Individual keratinocytes, particularly in the suprabasal layers, were uniformly flattened with widening of the intercellular spaces. Dermal capillaries were dilated and filled with erythrocytes (Fig. 4). The dermis was occupied by a dense infiltrate composed largely of plasma cells and lymphocytes with a slight admixture of mast cells, fibroblasts, and eosinophils (Fig. 5). In some areas, increased fibrosis and decreased amounts of elastic fibers (confirmed with elastic stain) were also noted. A prussian blue staining revealed no hemosiderin deposition.

Using a peroxidase-antiperoxidase (PAP) method applied on paraffin-embedded sections, we determined the immunoglobulin class distribution in the plasma cellular infiltrate. In all lesion, IgG-producing plasma cells predominated and IgA and IgM-positive cells were present in very low numbers. κ and λ light chain positive cells were present in equal quantities (Table 1).

| Composition of contents of plasma cells by peroxidase-antiperoxidase stain |
|-----------------|----------------|----------------|--------|--------|
|                 | IgG | IgA | IgM | κ     | λ    |
| 4+              | +   | +   | +   | 3+    | 3+   |
| −: (5% + : 5%−10% 2+ : 20−40% 3+: 40−60% 4+: 60%)

Electron microscopic examination showed vacuolar changes of cytoplasm and focal clumpings of tonofilaments in the keratinocytes with widening of intercellular spaces (Fig. 6). The infiltrate in the dermis was composed largely of plasma cells, which showed extensive well-developed rough endoplasmic reticulum with disruption of mitochondrial cristae (Fig. 7). No viral particles or elastic fibers were seen in the specimens.

As a therapeutic trial, he underwent a circumcision in January 1991. When he was last seen in March 1991, his prepuce lesion had almost completely resolved and he no longer complained of a squeezing pain on erection. But the lesions of glans penis seemed to be unchanged from his previous examination.

**DISCUSSION**

BCP is an intriguing disorder characterized by chronic erosive lesions of glans penis and prepuce, which resembles erythrolaspsia of Queyrat. Although some investigators noted that the lesion of BCP is shiny and moist rather than velvety and granular as in erythrolaspsia of Queyrat, it is generally believed that these
Fig. 1. Shallow ovoid erosion and surrounding erythema on the inner surface of the prepuce.

Fig. 2. Shiny glazed macular erythema on the glans penis and prepuce.

Fig. 3. Erythematous constricting band on the inner surface of the prepuce encircling the penile shaft.

Fig. 4. Biopsy specimen obtained from the erythematous constricting band on the inner surface of the prepuce shows a thin epidermis with a focal absence of horny and granular layer. Individual keratinocytes are flattened with widening of the intercellular spaces. Dermal capillaries are dilated and filled with erythrocytes (H & E stain, ×200).

Fig. 5. Photomicrograph of the dermal infiltrate composed largely of plasma cells and lymphocytes (H & E stain, ×200).

Two conditions are indistinguishable clinically. The only definitive method of establishing a diagnosis is by characteristic histologic changes affecting the epidermis and dermis. The epidermis is composed of diamond-shaped, flattened keratinocytes that are separated from each other by uniform intercellular edema. In addition to a characteristic band-like plasma cell infiltration in the dermis, the dermal blood vessels are dilated and there may be extravasation of erythrocytes and deposition of hemosiderin.
The most interesting aspect of our case was the clinical finding that the erythematous erosive lesion of the inner surface of the prepuce progressed to from a constricting band. This clinical feature could suggest a diagnosis of balanitis xerotica obliterans, which is considered as an end-stage condition evolving from chronic non-specific balanoposthitis. However, the absence of the typical smooth ivory-white sclerotic scarring of the glans and prepuce, as well as the histopathological findings, allows us to rule out this diagnosis. In our case, microscopic examination showed the above mentioned characteristic histological changes, except the findings of hemosiderin deposition.

In the series of twenty patients with balanitis and vulvitis circumspecta plasmacellularis, reported by Souteyrand et al., they stated that hemosiderin deposition was frequently marked, but occasionally could not be found even in prussian blue stained specimens. Furthermore, they stated that, in some cases, a degree of fibrosis and loss of elastica with easily visible fibroblasts could be seen. It is noteworthy that the biopsy specimen obtained from the erythematous constricting band of the prepuce revealed increased fibrosis and decreased amount of elastic fibers which correlate well with our clinical observations.

As previously noted, little is known about the etiology of this condition. Chronic infection by Mycobacterium smegmatis has been postulated and heat, constant friction, and poor hygiene may be predisposing factors, since the lesion occurs exclusively in elderly uncircumcised men.

Toonstra and van Wicher determined the immunoglobulin class distribution in the plasma cellular infiltrate using an unlabelled antibody peroxidase-antiperoxidase method. In their study, the main immunoglobulin class proved to be IgG with lower numbers of IgA and IgD and no or very few IgM-producing cells. They stated that these findings are suggestive of a nonspecific polyclonal stimulation of B-cells, which might be caused by a persistant infection. We also observed similar findings. Recently, however, Nishimura et al. observed IgE as the main immunoglobulin class in the plasma cellular infiltrate and suggested that, although a specific allergen has not been indentified yet, this condition may be related to immediate hypersensi-
tivity.

In 1988, Morioka et al' reported a 74-year-old woman with vulvitis circumscripta plasmacellularis, which is believed to be the female counterpart of BCP, in whom the presence of herpes simplex antigen was verified by direct immunofluorescence staining. She was successfully treated with intraleisonal injection of interferon-α. However, we were unable to confirm this by immunofluorescence investigation or by electron microscopic study, which failed to demonstrate a virus in the lesion of BCP. We feel that the tumoral character of the lesion in their case makes this diagnosis somewhat suspect, as is resembled plas-macanthoma described by Ferrira-Marques and Ramos E Silva.

The plasmacytic nature of the infiltrate was particularly stressed, giving rise to the commonly used terminology, BCP or plasma cell balanitis. In some cases, however, there is only a moderate or small number of plasma cells. Furthermore, it has been pointed out that plasma cells frequently predominate in the inflammatory response at the mucocutaneous junctions in a variety of benign and malignant process. Thus the terms, plasmacytosis circumferentialis, plasmacytosis mucosae, and plasma cell orificial mucositis were introduced for grouping these anatomic variants.

We hope that more extensive clinical descriptions of subsequently reported cases and further detailed studies, including immunohistochemical and electron microscopic findings, will better define this entity and permit clarification of its relationship to other diseases in this group.

REFERENCES


