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Balanitis xerotica obliterans – a review

Abstract *Balanitis xerotica obliterans* (BXO) is a scarcely known disease, wrongly considered rare. With a high degree of suspicion and histologic examination, the condition will prove to be much more frequent than one generally believes. The etiology of the condition is unknown at present. Many cases of BXO occurring after circumcision may be cases of secondary phimosis due to BXO not being recognized at the time of surgery. Most of the cases of BXO are seen in the third to fifth decades of life, even though they may occur at the extremes of age. Biopsy of the lesions is not essential in all cases and is indicated to differentiate from penile cancer and in atypical cases. Early diagnosis and treatment of BXO are very important in preventing the urological complications of the diseases such as urethral stricture. Treatment of BXO depends on the anatomic location of the lesions and their extent and severity, together with the rapidity of progression of the disease process. The treatment may vary from topical corticosteroids, laser vaporization in early cases to meatoplasty and urethroplasty in extensive cases. Topical pharmacotherapy is useful in the early stages to reduce the initial symptoms and slow down the progression, but is not effective in all cases and is not the curative treatment of disease. Meatal stenosis, phimosis, scar adhesions, fissures, erosions of glans and prepuce and involvement of the urethra are indications for surgical treatment. Surgery seems to be the only treatment that can relieve the symptoms of advanced disease. Modified circumcision, with total removal of inner preputial layer, definitively relieves phimosis without any recurrence. Meatotomy will not prevent the recurrence of meatal stenosis. Excision of the scleroatrophic tract and grafting of the glans base, coronal sulcus, and the end of the shaft give a complete relief of pain during erection and intercourse in circumcised patients with balanopreputial adhesions and restore the elasticity of the skin of penile shaft. These procedures have been shown to yield excellent functional results during a follow-up period of up to 4 years. BXO involving anterior urethra can be treated by 2-stage urethroplasty or substitution urethroplasty. The complete excision of the stricture and flap urethroplasty seems to be better than a 2-stage procedure. However, at the present time, it is not possible to say that surgery can completely resolve this chronic and progressive disease. Despite many reports in the literature of cases of BXO associated with squamous cell carcinoma, the etiologic relationship between the two conditions is uncertain.

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Introduction

The term balanitis xerotica obliterans (BXO) was first coined by Stuhmer in 1928, for the chronic, progressive scleroatrophic inflammatory process of unknown etiology affecting the glans penis, prepuce, and urethral meatus. The lesions occur as plaques or papules on the prepuce or glans penis and may extend to result in urethral meatal stenosis. BXO has 3 components: '*balanitis*', meaning chronic inflammation of the glans penis; '*xerotica*', meaning an abnormally dry appearance of the lesion; and '*obliterans*', for the association of occasional endarteritis [1]. Fruhwald [2] and Grutz [3] have reported penile carcinoma in association with BXO.

Freeman et al. (1941) have reported on 18 cases of BXO and are of the opinion that BXO is a disease of insidious onset with slow progression occurring at any age and not related to circumcision [4].

Clinical Features

BXO is a disease of unknown etiology affecting the prepuce, glans, and often the urethral meatus. BXO involving the anterior urethra and membranous urethra has also been reported [5].

The disease starts insidiously. There may be associated pain, pruritus and 'burning' or 'pricking' sensation. Rarely, purulent urethral discharge may be present. The lesions may present in 3 forms: (1) typical ivory-white macules; (2) confluent plaques or hemorrhagic bullae on the glans following sexual intercourse; and (3) meatal and perimeatal lesions combined with difficulty in voiding. Even though BXO is a chronic and progressive disease, it sometimes takes a cyclical course with periods of remission in which the genital skin becomes less atrophic and the phimosis and meatal stenosis become less severe, with reduction in subjective symptoms [6].

The prepuce is involved in uncircumcised patients in the form of a sclerotic, constricting band about 1–2 cm from the distal end, which may progress to phimosis. Glans may be involved in a diffuse fashion or as mottled patches. There may be a whitish discoloration of the perimeatal region of the glans penis (Fig. 1). A combination of white plaque and normal red tissue may give a mosaic appearance to the lesion. Sometimes telangiectasia and hemorrhagic petechiae have also been reported [6]. The pathognomonic feature of BXO includes a perimeatal whitish and erythematous area. Gradually the acute condition becomes chronic and may present as a "burn out" lesion.

Montgomery and Hill reported that there is no association between lichen sclerosus et atrophicus (LSEA) and BXO [7]. Laymon and Freeman reported on 24 cases of BXO and felt that LSEA and BXO were identical and that the former could occur on the genitals as isolated papules on the shaft or as sclerotic preputial patches with urethral involvement. These authors consider that BXO is the male genital form of LSEA. This view is held by many even today [6]. At present, the evidence is inconclusive regarding whether BXO is a localized form of LSEA, which occurs on the upper trunk, clavicular and scapular regions, neck, axillae, and forearms as flat-topped irregular white or yellowish-white papules. Laymon and Freeman et al. found patches of lichen sclerosus on the trunk and limbs in four of six patients with BXO [8]. However, none of the four patients with BXO described by Staff had patches of lichen sclerosus [9]. In a series of eight male cases ofLSEA, changes on the genitalia were found in only one, with lesions on the shaft of penis, the prepuce, and glans [10].

The disease most often occurs in patients 30–49 years of age, though it has been reported in adolescents (8–10 years) as well as in extremes of age [5]. The patients may present with primary or secondary phimosis, dysuria, and obstructive voiding symptoms. Bainbridge et al. reported some degree of urinary obstruction in 47% of patients in their series of 17 [11]. Three of their patients had a meatal stenosis and three had, in addition, a urethral stricture which caused an outflow obstruction.

Occasionally, other fibrosing or synechial pathological entities such as mucosal pemphigoid or chronic balanoposthitis may lead to a clinical picture similar to that of BXO.

In the advanced stages of the disease, the coronal ridge can be partially or completely obliterated due to balanopreputial adhesions, and the frenulum can become sclerotic, so much so that it is spontaneously re-

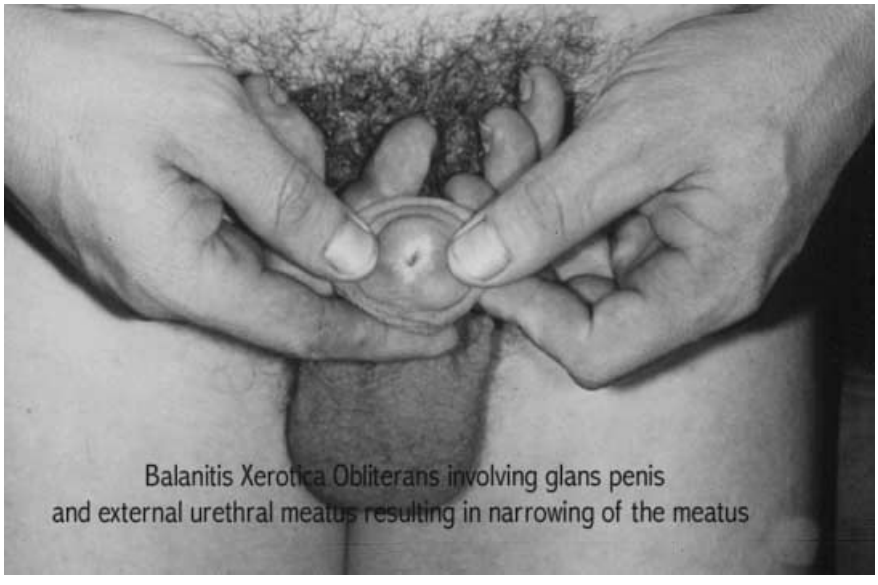

Fig. 1 Balanitis xerotica obliterans involving glans penis and external urethral meatus resulting in narrowing of the meatus

tracted and may even disappear. Catterall and Oates described a perimeatal collar of white fibrous tissue involving the external meatus and extending into the anterior urethra [12]. It is not often realized that, in certain cases, the urethral involvement may be quite extensive. This poses problems, not only of management, but also of diagnosis, if the condition is not recognized.

Urethral involvement by BXO was first reported in 30 patients by Laymon in 1951 [13], including some with meatal stricture. Their main symptoms included pain, irritation, and disturbance of sexual function [11]. Penile urethral involvement by BXO has also been reported by Catterall et al. and Staff [9, 12].

Catterall and Oates reported that the main symptom of patients with BXO is urethral discharge [12]. One third of their patients also had urinary problems, such as dysuria and obstructive voiding symptoms. Urethral involvement was in the form of meatal lesions with extension into the squamous epithelium of fossa navicularis. Some of their patient also had white plaques in the anterior urethra.

Diagnosis

The diagnosis of BXO cannot be made on clinical grounds alone, since the BXO lesions resemble *erythroplasia of Queyrat*, lichen planus, leukoplakia, and scleroderma. However, the histological features of BXO are distinct and specific. Biopsy will therefore be extremely useful in diagnosis in these circumstances.

BXO and circumcision

BXO can occur following circumcision [1], but further reports have failed to establish this as a causative factor [12]. BXO following partial amputation of the penis has been reported [14, 15]. It can also occur before the onset of penile cancer [16]. Even though chronic inflammation and phimosis are established etiological factors for carcinoma of the penis, their etiological role in BXO has not been settled so far [6].

Histopathology

Histologically, BXO is essentially the same as LSEA [8]. Both are characterized by hyperkeratosis of the epithelium, hydropic degeneration of the basal cells, sclerosis of the subepithelial collagen, dermal lymphocytic infiltration, atrophic epidermis with loss of rete pegs, homogenization of the collagen in the upper third of the dermis, and a more or less dense zone of lymphocytes and histiocytes beneath the homogenized collagen (Figs. 2, 3). The small arteries and arterioles of the upper and middle dermis may show evidence of endarteritis obliterans, but this is not a consistent feature.

Datta et al. (1993) suggested an association of autoimmune diseases like vitiligo is responsible for BXO [17]. Immunoglobulin studies showed IgG to be increased, which indicates an antigenic stimulation [17]. This idea was promulgated due to the fact that skin exposed to urine in non-physiological areas, e.g., around periurethral fistula or around suprapubic cystostomy, manifests alterations similar to those caused by BXO. Circumcision exposes the glanular mucosa and adjacent urethra to the antigenic stimulation, leading to the histological changes of BXO. IgM has also been shown to increase, which may be due to the coexistent chronic infection, arthritis, and vasculitis, all of which may be autoimmune in nature or may be due to BXO itself.

Hinchliffe et al. (1994) feel that limited immunophenotyping may be a useful adjunct to the diagnosis of BXO in pediatric cases in which only limited tissue is available [18]. These authors have shown that the infiltrate in BXO patients was wholly composed of T cells (positive with UCLH-1 antibody) in all cases. B cells (positive with L-26 antibody) were found only focally in small, discrete, easily recognizable (follicular or early follicle-like) aggregates, positioned slightly deeper than the band-like infiltrate of T cells. T cells were inconspicuous in nine of the 12 control specimens. In the three other controls, T cells were much more obvious and these patients showed clinical features suggestive of BXO. Increased IL-6 in the epidermis of the prepuce may be related to the inflammatory pathophysiology of BXO [19].

Decrease of the elastic tissue in the superficial layer is also a common histological finding due to the loss of antigenicity of macromolecules in the homogenized zone of collagen. All these observations suggest that BXO may result from an autoimmune process [17].

Clinical management

The traditional treatment for BXO has been dilatation of the urethra when stenosis occurs, plus the use of topical emollients. Topical corticosteroid therapy is useful in the early phase and lessens the initial symptoms and slows the progression of the disease process. A partial resolution of the clinical picture has been observed over periods of up to 3 years [6]. This type of local treatment is not uniformly effective. Topical steroid therapy is used after meatal stenosis has occurred and following urethral dilatations.

Localized BXO on prepuce and glans can be treated with mild topical corticosteroids such as 1% hydrocortisone over a 6-week period with a gradual reduction in the number of applications. The corticosteroids control nonspecific inflammation of the glans and preputial sac. For protracted periods (1–2 months), the treatment consists of 0.1% cream of betamethasone-17 valerate together with emollient ointment, and this dosage is repeated many times during the course of treatment.

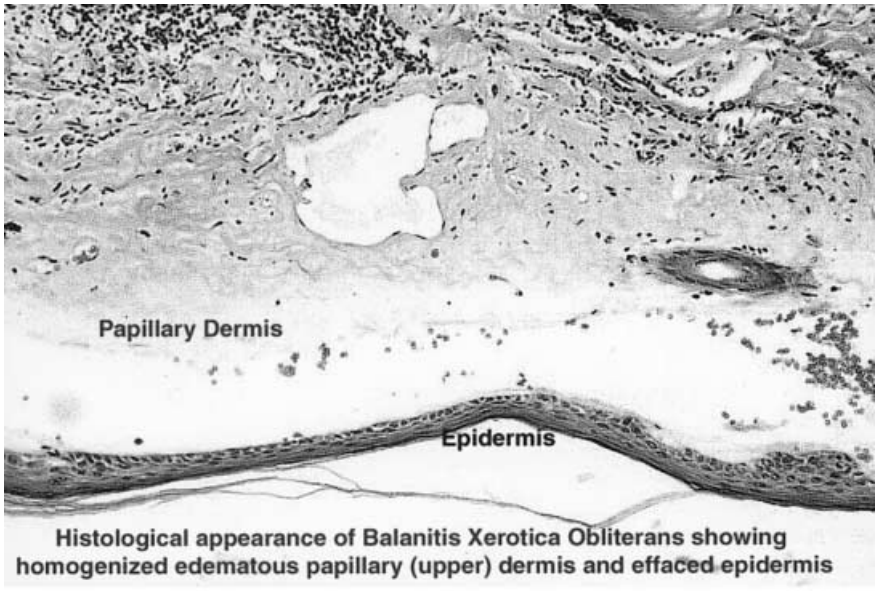

Fig. 2 Histological appearance of early stage balanitis xerotica obliterans showing homogenized edematous papillary (upper) dermis and effaced epidermis (hematoxylin and eosin, ×20)

Histological appearance of Balanitis Xerotica Obliterans showing homogenized edematous papillary (upper) dermis and effaced epidermis

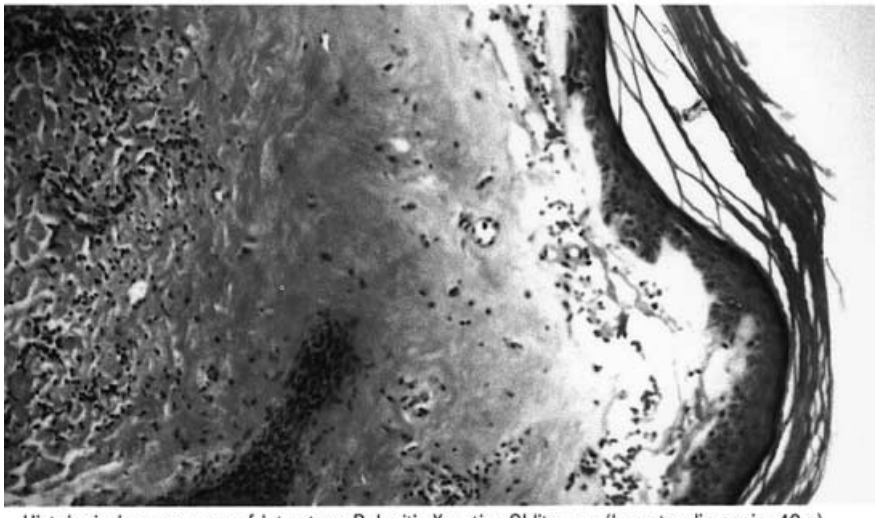

Fig. 3 Histological appearance of late stage balanitis xerotica obliterans (hematoxylin and eosin, ×40) showing less edema in the upper dermis and more sclerosis throughout the dermis. Involvement of the lower dermis and fat is also seen

Histological appearance of late stage Balanitis Xerotica Obliterans (hematoxylin eosin, 40 x) showing less edema in the upper dermis and more sclerosis thoughout the dermis. Involvement of the lower dermis and fat is also seen

Potent steroid applications such as betamethasone valerate 0.1% should be avoided, especially in pediatric patients [6]. The duration of treatment and severity of the disease process influence the observed response to the topical steroid therapy.

Triamcinolone acetonide in a dose of 10 mg/ml combined with xylocaine in a final concentration of 5 mg/ml can be used for intralesional injection and will reduce the pruritus and burning with decreased preputial sclerosis and thickening [20]. Xylocaine will reduce the discomfort resulting from triamcinolone injection.

Recently, continuous wave carbon dioxide (CW-CO_2) laser vaporization has been successfully used in the treatment of BXO. Laser energy will enable an accurate control of the beam, minimizing damage to the surrounding tissue, which is possible because of the ability of laser treatment to predict and control the depth of surgical injury by alterations in power density and time characteristics. The lesions of BXO will completely disappear if there is an absence of an associated involvement of meatus and/or urethra. Re-epithelialization takes place by 6 days after laser therapy of the lesions [21–23].

Surgical treatment

In some patients, topical therapy produces only a poor and transitory alleviation of symptoms during the initial period of therapy; then the disease becomes slowly progressive and worse. In such cases with a longer duration of the disease process, surgical treatment is mandatory.

Urethral dilatation by bougienage alone will be adequate for meatal stenosis with nonspecific inflam-

matory changes. In severe cases, onlay flap/2-stage urethroplasty will be necessary, together with a complete excision of the involved portion of urethra [21]. Urethral meatal stenosis can be treated by ventral meatotomy or dorsal V-meatoplasty. Meatotomy in BXO is often followed by restenosis, so that the interposition techniques with normal skin flap (substitution urethroplasty) are advised [24]. Surgical correction of the meatus, however, does not improve the common loss of sensitivity in the glans penis. Zungri et al. [24] reported that a complete resection of the glans mucosa and meatoplasty produced complete resolution of this disease in their case, allowing the normal epithelium to grow over and cover the glans.

Modified circumcision with total removal of the inner preputial skin is performed as described by Jaenner et al. [25]. In this procedure, a circular incision is made on the external preputial surface below the inferior edge of the sclerotic part, generally 1–1.5 cm from the apex. The skin and subcutaneous tissues are undermined, and the two preputial layers are separated. The glans is well exposed by means of a dorsal vertical incision of the internal layer. Then the distal tract and the whole inner surface of the foreskin are cut away by a circular incision just below the glans base. The upper edge of the remaining portion of the external preputial layer is sewn up all around the glans base with interrupted sutures.

In previously circumcised patients with balanopreputial adhesions, the scleroatrophic tract intersecting the skin of the glans base and the residual prepuce are completely excised [26]. Full-thickness skin grafts (1–1.5 cm in width) can then be placed in a circular fashion, covering the glans base and distal portion of the penile shaft. The grafts are sutured by interrupted catgut sutures and fastened by elastic dressing. No urethral catheters or urinary diversions are necessary following these procedures. These procedures yield good results without recurrence of phimosis, meatal stenosis, balanopreputial adhesions, pain during erection, or worsening of glandular sclerosis, and without retraction of the grafts and affecting the penile skin or the remaining external preputial skin [26].

Herschorn et al. reported on a case of biopsy-proven BXO involving the penile urethra as far back as the penoscrotal junction occurring in a patient with a long history of urethral stricture and many urethroplasty failures [27]. Of more interest, in this patient, squamous cell carcinoma involving the external urethral meatus and fossa navicularis eventually developed following the diagnosis of BXO.

BXO involving anterior urethra can be treated by 2-stage urethroplasty or substitution urethroplasty. Flap urethroplasty seems to be better than a 2-stage procedure as the former involves a complete excision of the involved urethra with reconstruction in one or two stages, using a flap or steroid injection into the area after first-stage urethroplasty to obtain regression of the disease prior to the 2nd stage closure [27].

Conclusions

BXO is a chronic inflammatory dermatosis of as yet unknown etiology involving the male external genital areas (external urethral meatus, prepuce, anterior urethra in the male) and results in the formation of white plaques with epidermal atrophy. This may be a local male genital form of LSEA. Inflammation and altered fibroblast function in the papillary dermis leads to fibrosis of the upper dermis. Local irritation, infection by uncommon organisms (e.g., *Borrelia*), and genetic and autoimmune factors have been suggested to have possible etiological roles.

Genital skin and mucosa are most frequently affected, but extragenital lesions do occur, and even rare oral presentations are reported. Involvement of urethra, penile shaft, and scrotum is rare. Genital presentations outnumber extragenital reports by more than 5:1. The condition should be differentiated from Bowen's disease, leukoplakia, lichen planus, and other conditions involving the prepuce and glans penis. Skin biopsy is helpful in the diagnosis and shows infiltrate in the dermal–epidermal junction, compact hyperkeratosis with stratum corneum often thicker than the effaced epidermis. In late stages, the edema in the papillary (upper) dermis is replaced by a dense, homogeneous fibrosis.

The condition commonly presents with dysuria, glanular/preputial itching (mild cases), and urinary obstruction due to phimosis, meatal stenosis, or anterior urethral stricture (late stages). Urethral involvement should be excluded by the appropriate investigations (retrograde urethrogram) in extensive lesions involving prepuce, glans penis, and external urethral meatus. There may be an increased risk of squamous cell carcinoma in late stages.

Early lesions respond to such local measures as the application of topical steroids (betamethasone, triamcinolone, clobetasol, isotretinoin) and laser vaporization. Meatal stenosis is managed by meatotomy or meatoplasty (and modifications thereof, e.g., dorsal 'v' meatoplasty). Extensive urethral involvement should be managed by substitution urethroplasty using skin or buccal mucosa.

Overall prognosis is good if the condition is diagnosed early and such sequelae as meatal stenosis and urethral stricture are appropriately managed.

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