

# Treatment of balanitis xerotica obliterans in pediatric patients

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## ABSTRACT

**Objectives.** Balanitis xerotica obliterans (BXO) is a chronic inflammatory disease with a little known incidence in pediatric population. The objective of this work was to describe our experience in the treatment of BXO.

**Materials and methods.** Retrospective study carried out in 419 patients undergoing circumcision surgery between January 2014 and January 2017. Demographic, clinical, therapeutic, and anatomical and pathological variables, as well as complications during follow-up, were analyzed.

**Results.** Of the 419 patients, 41 (9.78%) were diagnosed with BXO. 6 patients were excluded owing to lack of follow-up, so 35 patients were analyzed.

Mean age at diagnosis was 8.6 years. Suspicion diagnosis was clinical at physical exploration in 17 patients (48.6%), and at surgery in 18 patients (51.4%). Anatomical and pathological confirmation was performed in a total 35 patients (100%).

During follow-up, 6 patients (17.14%) had lesions in the glans, 3 (8.57%) in the urethra, and 9 (25.71%) in both. 6 meatotomies (17.14%) and 5 new circumcisions (14.28%) had to be carried out. Mean recurrence time was 32.43 months. In 19 patients (54.28%), topical corticoids – ointment – were applied, and 1 patient (2.85%) received topical immunosuppressants.

**Conclusions.** A close follow-up of patients with clinical or anatomical and pathological diagnosis of BXO is required given its high morbidity. The complications described in pediatric population include meatal and urethral stenosis, as well as recurrent phimosis, unless a sufficient amount of foreskin is resected.

**KEY WORDS:** Balanitis xerotica obliterans; Treatment; Complications; Pediatrics.

## TRATAMIENTO DE LA BALANITIS XERÓTICA OBLITERANTE EN PACIENTES PEDIÁTRICOS

### RESUMEN

**Objetivos.** La balanitis xerótica obliterante (BXO) es una enfermedad crónica inflamatoria de incidencia poco conocida en la población pediátrica. El objetivo de este trabajo es describir nuestra experiencia en el tratamiento de las BXO.

**Material y métodos.** Estudio retrospectivo de 419 pacientes intervenidos de circuncisión en el periodo comprendido entre enero de 2014 y enero de 2017. Analizamos variables demográficas, clínicas, anatomopatológicas, terapéuticas y complicaciones durante el seguimiento.

**Resultados.** De los 419 pacientes, 41 fueron diagnosticados de BXO (9,78%). Seis pacientes fueron excluidos por falta de seguimiento, por lo que se analizaron 35 pacientes.

La media de edad al diagnóstico fue de 8,6 años. El diagnóstico de sospecha fue clínico durante la exploración física en 17 pacientes (48,6%) y durante la intervención en 18 (51,4%), realizando la confirmación anatomopatológica en un total de 35 pacientes (100%).

Durante el seguimiento 6 pacientes (17,14%) presentaron lesiones en glándula, 3 (8,57%) en uretra y 9 (25,71%) en ambas localizaciones, siendo necesaria la realización de 6 meatotomías (17,14%) y de nueva circuncisión en 5 (14,28%). El tiempo medio de recidiva fue de 32,43 meses. En 19 pacientes (54,28%) se aplicaron corticoides tópicos en pomada y en 1 paciente inmunosupresores tópicos (2,85%).

**Conclusiones.** Es necesario un seguimiento estrecho de los pacientes con diagnóstico clínico o anatomopatológico de BXO dada su elevada morbilidad. Las principales complicaciones descritas en la población pediátrica son la estenosis meatal y uretral, así como la recidiva de la fimosis si no se reseca el prepucio suficientemente.

**PALABRAS CLAVE:** Balanitis xerótica obliterante; Tratamiento; Complicaciones; Pediatría.

## INTRODUCTION

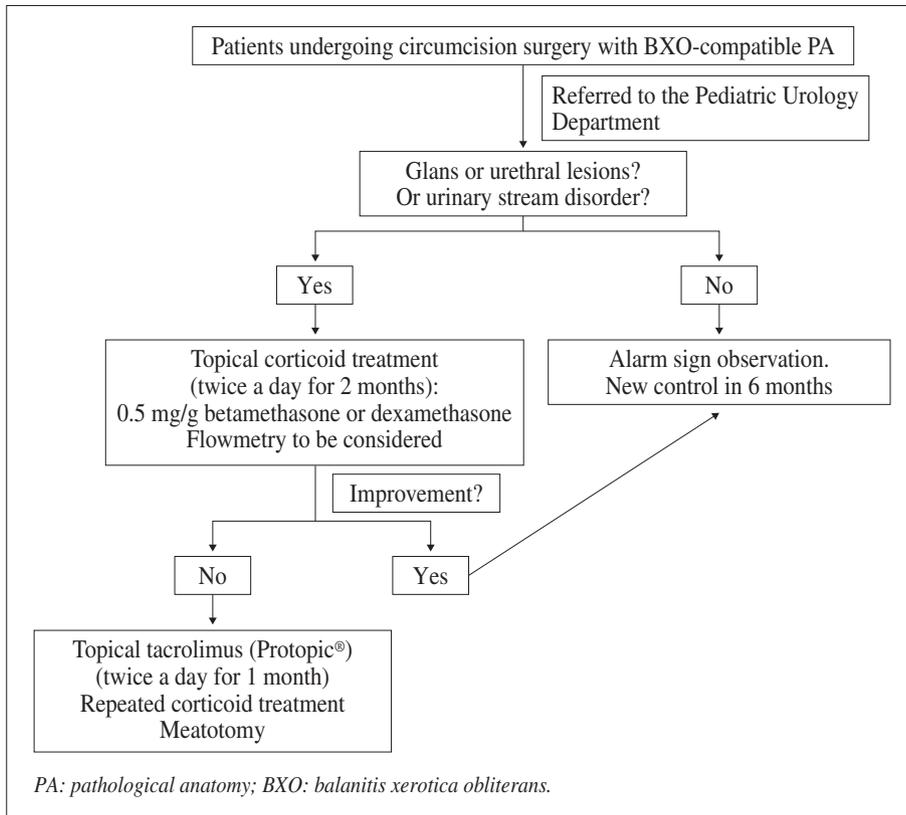
Balanitis xerotica obliterans (BXO) is a chronic inflammatory disease (male variant of lichen sclerosus) that is rare in pediatric population. It can involve not only the penis, but also the foreskin, the glans, the meatus, and/or the urethra. Its actual incidence is unknown, since there

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**Figure 1.** Follow-up protocol in patients diagnosed with BXO.

are no publications submitting all foreskins for anatomical and pathological study following circumcision<sup>(1)</sup>. In phimosis children, incidence is estimated at 5-52%, and it has been described even in neonatal patients<sup>(2,3)</sup>. The degree of clinical suspicion has risen as a result of having a better knowledge of the condition, which in turn has caused an increase in incidence records in the last decades.

Diagnosis is based on clinical suspicion and is confirmed with the histopathological study of the foreskin<sup>(1,4)</sup>. The most frequent complications associated with BXO include recurrent phimosis (especially in case of incomplete foreskin resection) and meatal or urethral stenosis, which can occur in up to 47% of patients. Recurrence has been recently reported in up to 40% of patients with hypospadias treated with two-stage oral mucosal graft. In adult patients, it is associated with penile squamous carcinoma, with an estimated incidence of around 20%<sup>(5-7)</sup>.

The objective of this work was to analyze our experience in the treatment of pediatric patients with BXO. Medical treatment success rate and related complications, as well as their treatment, were analyzed.

## MATERIALS AND METHODS

A retrospective study was carried out in 419 patients undergoing circumcision surgery in our healthcare facility between January 2014 and January 2017.

Data on demographic variables, initial and follow-up physical exploration, diagnostic methods, initial treatment, and follow-up complications and their treatment were analyzed.

## Diagnosis

In our healthcare facility, the anatomical and pathological study of the foreskin is not routinely carried out in patients undergoing phimosis surgery, unless BXO is clinically suspected – either preoperatively at external consultations, or at surgery.

If BXO is clinically suspected, the foreskin is fully resected at circumcision. If the anatomical and pathological study of the foreskin sample allows the condition to be confirmed, the patient is referred to external pediatric urology consultations for outpatient follow-up and early complication detection.

## Initial postsurgical treatment

If macroscopic lesions are found in the glans or the urethra, topical corticoid treatment is initiated (0.5 mg/g betamethasone or dexamethasone every 12 hours for two months), and flowmetry is considered. If no improvement is noted in spite of topical treatment, the cycle is repeated, topical tacrolimus treatment is applied (every 12 hours at 0.03% for one month). In patients with meatal stenosis refractory to medical treatment, meatotomy is indicated (Fig. 1).

**Table 1. Description of the macroscopic lesions found in BXO patients during quarterly outpatient follow-up.**

Outpatient follow-up Controls (N)	Location			New lesions Glans
	Glans	Urethra	Both	
1 <sup>st</sup> control (N:18)	6 (33.3%)	3 (16.7%)	9 (50%)	–
2 <sup>nd</sup> control (N:11)	3 (37.5%)	2 (25%)	3 (27.7%)	2 (18%)
3 <sup>rd</sup> control (N:9)	5 (55.6%)	2 (22.2%)	2 (22.2%)	–

*BXO: balanitis xerotica obliterans.*

**Follow-up**

In patients without macroscopic lesions, outpatient controls are carried out every 6 months while explaining the rules for new consultations – occurrence of macroscopic lesions and voiding disorders primarily.

In patients with macroscopic lesions, outpatient controls are carried out every two months to assess medical treatment efficacy and control complication occurrence.

Flowmetry is indicated in patients with urinary stream disorders or need for abdominal press when voiding – assessed at each individual consultation. The occurrence of flowmetry disorders – especially plateau curve – or voiding difficulty are indications for meatotomy.

All patients are controlled at external urology consultations up to 16 years of age, and subsequently referred to their local urologist.

**RESULTS**

In the study period, 419 patients underwent circumcision surgery, 41 (9.78%) of them being diagnosed with BXO. 6 patients were excluded from the study owing to lack of follow-up, so there were 35 (8.78%) BXO patients eligible for analysis.

Mean age at diagnosis was 8.6 years (range: 4-14 years), and the main indication of the procedure was progressive phimosis in all patients (100%). However, 2 patients had meatal stenosis at diagnosis.

BXO incidence in circumcised patients increased progressively with study years, with 8 (5.3%) patients in the first year, 13 (9.15%) patients in the second year, and 14 (11.2%) patients in the third year.

**Initial diagnosis**

Suspicion diagnosis was clinical at preoperative physical exploration in 17 (48.6%) patients, and intraoperatively in 18 (51.4%) patients. 5 patients who had previously undergone circumcision with partial foreskin resection had recurrent phimosis, which raised suspicion of BXO.

**Confirmation diagnosis**

Anatomical and pathological confirmation was performed in the 35 patients (100%), demonstrating epidermal

atrophy with collagenization and histological and inflammatory infiltration.

**Initial treatment**

Circumcision with complete preputial skin resection was carried out in all patients (100%). The 2 meatal stenosis patients also underwent meatotomy in the same surgical procedure (5.7%).

**Follow-up**

Outpatient follow-up was performed in all patients at pediatric urology external consultations 3 weeks (range: 1-4 weeks) following the procedure.

In the first outpatient control, 18 patients (51.4%) had macroscopic penile lesions: 6 (17.1%) lesions in the glans, 3 (8.57%) lesions in the urethra, and 9 (25.71%) lesions in both locations.

Of the patients with macroscopic lesions, 17 (94.4%) patients initially received topical corticoid treatment, 1 (5.5%) patient received tacrolimus treatment, and 1 (5.5%) meatotomy was required. Topical treatment was initiated at BXO confirmation in the anatomical and pathological study.

In subsequent controls, the macroscopic lesions disappeared with treatment in 12 (66.7%) patients, and new lesions appeared in 2 (11.1%) patients (Table 1).

Today, 9 patients have macroscopic lesions, 5 (14.2%) of whom are receiving topical corticoid treatment, and 4 (11.4%) of whom are receiving tacrolimus treatment (Table 2).

Of the 35 patients, urinary stream disorders were noted in 12 (34.3%) patients – mild disorders in 8 (22.9%) patients, and moderate disorders in 4 (11.4%) patients. All of them had macroscopic disorders in the urethral meatus.

**Complications**

4 (11.4%) patients had urethral meatus stenosis during the first 9 months of follow-up (range: 1-9 months). All of them required meatotomy.

Mean follow-up was 32.4 months (SD: 4).

**DISCUSSION**

BXO is a dermatologic condition involving the male genital organs. It was first described by Stühmer et al.

**Table 2. Treatment description in BXO patients during outpatient follow-up.**

<i>Outpatient controls</i>		<i>1<sup>st</sup></i>	<i>2<sup>nd</sup></i>	<i>3<sup>rd</sup></i>
Medical treatment	Corticoids	17 (48.5%)	5 (14.2%)	5 (14.2%)
	Tacrolimus	1 (2.85%)	3 (8.5%)	4 (11.4%)
Surgical treatment	Meatotomy	1 (2.85%)	1 (2.85%)	2 (5.7%)

*BXO: balanitis xerotica obliterans.*

in 1928, and it is regarded as the male variant of lichen sclerosus<sup>(8)</sup>.

Today, BXO is one of the main causes of secondary and progressive phimosis in children. Etiology is multifactorial, with genetic, autoimmune, hormonal, and infectious factors<sup>(1,2)</sup>.

Incidence is estimated at 5-52% according to the series<sup>(3)</sup>, but actual incidence is underrated as many cases are healed with circumcision, and many foreskin samples are not submitted for histological and pathological analysis as a result of lack of knowledge regarding the pathology. In our series, an increase in incidence has been noted in the last years (up to 11.2% of circumcised patients in the last study year) owing to the fact that circumcision surgeons have a higher degree of clinical suspicion as a result of having a better knowledge of the condition.

Diagnosis is based on clinical suspicion, which usually demonstrates secondary and progressive phimosis associated with a xerotic glans and foreskin appearance. In urethral involvement cases, dysuria, abdominal press voiding, and urinary retention episodes may occur<sup>(9,10)</sup>. Histological examination confirm diagnosis and demonstrates hyperkeratosis and epidermal basal layer atrophy, with loss of elastic fibers and collagen disorders associated with inflammatory infiltration. Clinical correlation with the anatomical and pathological study is variable, ranging between 2 and 88%<sup>(11)</sup>. In our series, clinical correlation was 100%.

There are no preventive measures for this pathology. Treatment is focused on limiting progression and detecting complications, which makes early diagnosis key.

There seems to be a certain consensus in the literature regarding first-line treatment in all cases – circumcision with complete foreskin resection. However, there are recent articles on the use of topical corticoid treatment to avoid circumcision, with success rates of 34.8%<sup>(12)</sup>.

Adjuvant treatments such as topical corticoids and immunosuppressants can be administered pre- or post-surgery, and as a maintenance treatment. Corticoids are the most widely used agents as they are more effective than tacrolimus, according to the literature<sup>(9)</sup>. In our experience, topical betamethasone (0.5 mg/g doses every 12 hours) reduces the prevalence of penile mac-

roscopic lesions by 70% (12/17), vs. 60% according to other series<sup>(7)</sup>.

Immunosuppressants such as topical tacrolimus (0.1%) have demonstrated to be a safe, well-tolerated treatment following circumcision. They are primarily used in patients with macroscopic lesions in the glans or the meatus, with a 9% recurrence rate<sup>(13)</sup>. In our healthcare facility, topical tacrolimus (at 0.03% doses every 12 hours) is only used in case of recurrence or persistence of macroscopic lesions in spite of topical corticoid treatment administration. Tacrolimus was only used from baseline in one patient, with poor results, as he still has macroscopic lesions in the glans.

Other treatments described include the administration of hydrocortisone or mometasone (at 0.05% doses), with a better result in terms of preputial skin retractability, and a poorer result in terms of inflammation degree in foreskin biopsies as compared to placebo<sup>(14)</sup>.

Triamcinolone – via an intraoperative intralesional injection at preputioplasty – has also been used, with a success rate of up to 81% in some series published<sup>(15)</sup>.

An adequate follow-up of these patients should be carried out in order to diagnose complications early and schedule the most suitable treatment. Flowmetry has demonstrated a high sensitivity in the early diagnosis of urethral stenosis<sup>(16)</sup>. However, this procedure is not routinely carried out in our healthcare facility, except in case of voiding disorders.

The most frequent complications of BXO include meatal stenosis and recurrent phimosis. Meatal and urethral involvement in BXO can cause severe clinical problems, such as urinary obstruction<sup>(16,17)</sup>. In urethral involvement cases with no response to topical treatment, meatotomy can prove necessary – in case of distal involvement –, and even complete urethral resection and replacement with another tissue such as the oral mucosa – in case of complete urethral involvement – may be required<sup>(7)</sup>. Our percentage of meatotomies (17.1% in total) is lower than in other series described in the literature – 47%<sup>(18)</sup>.

Up until now, no penile squamous carcinoma cases have been described in children with BXO, but there are few long-term follow-up data. Today, 21% of penile cancer patients have history of BXO, which is regarded as a pre-malign lesion<sup>(19,20)</sup>. Therefore, a close follow-up of BXO patients should be carried out up to adult age.

## CONCLUSIONS

BXO incidence has increased in the last years, most likely due to a better knowledge of the disease. In most patients, evolution is benign, and the condition is adequately controlled using topical corticoids. However, up to 39% of patients have complications, namely urethral stenosis and recurrent phimosis.

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