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## Endourology

# Meatotomy and meatoplasty on meatal stenosis due to balanitis xerotica obliterans

Rizky Lukman Hakim<sup>a</sup>, Aninda Dinar Widiantari<sup>b,\*</sup>

- <sup>a</sup> Department of Urology, Sulianti Saroso Hospital, Jakarta, Indonesia
- <sup>b</sup> Working Team of Research, Sulianti Saroso Hospital, Jakarta, Indonesia

#### ARTICLE INFO

Keywords:
Balanitis xerotica obliterans
Meatal stenosis
Meatotomy
Meatoplasty

#### ABSTRACT

Balanitis xerotica obliterans (BXO) is a chronic inflammatory disease affecting the male genitalia that is mostly underdiagnosed and undertreatment. We presented a 40-year-old man with meatal stenosis and an inability to urinate. Discoloration at the tip of the penis was found, and he said it got wider as he grew up. We conducted meatotomy and meatoplasty to release obstruction. Early diagnosis is essential to avoid complications.

#### 1. Introduction

Balanitis xerotica obliterans (BXO) is a chronic inflammatory disease typically affecting the foreskin and glans. The incidence was not well reported and underdiagnosed. The prevalence of the disease is unclear but predicted to be approximately 1 %. We presented a 40-year-old man with meatal stenosis due to BXO who's experienced the disease since he was a child.  $^{1}$ 

#### 2. Case presentation

A 40-year-old was admitted to the emergency room due to the inability to urinate for one day. He also complained of decreasing flow urination for one year. Since he was a child, he mentioned the discolouration of the tip of the penis to whitish and got wider as he got older (Fig. 1). He worked as a businessman and has gotten married with one kid. He has been circumcised and has a normal body mass index. He also has no history of diabetes mellitus.

In physical examination, it was found *orificium urethra externum* (*OUE*) area and *glans penis* was whitish and had a pinpoint appearance of OUE (Fig. 2).

In the intraoperative procedure, an incision was made in the 12 and 6 o'clock directions, and the 16-french catheter was inserted. The catheter could be inserted until the bulbous urethra before it became resistant. Then it was put out before being inserted again using mandrain stylet, impenetrate resistance as bulbous urethral stricture until reaching inside the bladder and urine flows out from the catheter (Fig. 3). The catheter was left to support the urethra for two weeks (Fig. 4). The urethral

orifice was treated with topical antibiotics twice a day. We also prescribed 3rd-generation cephalosporin for oral antibiotics within 7 days.

The catheter was removed two weeks after surgery, and the patient could urinate normally. We put this patient in 6 months follow-up. There was no clinical appearance of narrowing of the urethral orifice until 6 months of observation, and there was no decrease in urinary flow or problem in ejaculation or erection based on the patient's complaint.

## 3. Discussion

Balanitis xerotica obliterans is a lichen sclerosis (LS) affecting the genital area of men, particularly in the foreskin and glans, and/or meatus.  $^{1-3}$  In Europe and the US, BXO is reported to be 0.06–0.4 per 1000 boys per year and is usually found in one—to sixteen-year-old boys, with an average age of 6.8. A previous study by Kiss et al. mentioned that 40 % of BXO cases are related to phimosis. Boksh and Patwa estimated its prevalence in the UK is 0.1–0.3 %.

Conducting surveillance of BXO is challenging, as the clinical presentation varies and affects a wide array of physician specialities.4 Several studies have shown no race predilection; however, Nguyen et al. report higher rates in African American and Hispanic populations, likely secondary to lower rates of circumcision. <sup>5</sup> In Indonesia itself, surveillance on BXO isn't available.

BXO is a rare and chronically inflammatory disease that has an unknown aetiology.  $^2$  LS is associated with genetic components in women and autoimmune diseases. BXO itself is related to atopic dermatitis in men. Its relation with the Epstein-Barr virus and HPV as causative agents is also not proven.  $^{1,6}$  However, some previous studies found that BXO

<sup>\*</sup> Corresponding author. Working Team of Research, Sulianti Saroso Hospital, Sunter Permai Raya St., North Jakarta, Jakarta, 14340, Indonesia.

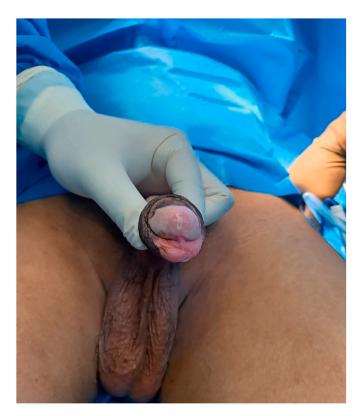


Fig. 1. The whitish appearance of the penile glans and nearly full closure of OUE.

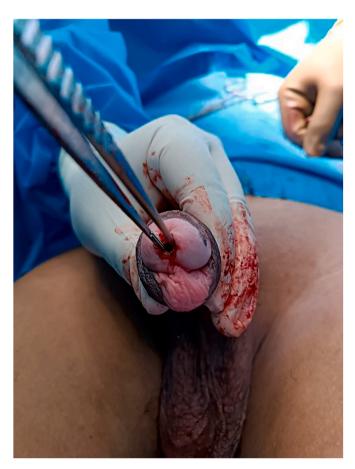


Fig. 2. The OUE before meatotomy.



Fig. 3. Post-meatoplasty appearance.

was associated with diabetes, HLA-DR11, HLA-DR12, and HLA-DQ7 antigen, and/or inflammatory causes.2,5,7 Post-micturition dribbling has been proposed as the pathogenesis of BXO. Also, in uncircumcised men, urine is more likely to pool between the prepuce and the glans penis, causing occlusion and precipitating the Koebner phenomenon and inflammation. Koebner phenomenon also describes the development of skin lesions along previous sites of trauma and irritation. Histologically, BXO lesion is characterised by an abundance of infiltrating, auto-reactive cytotoxic T-lymphocytes, impaired extracellular matrix metabolism and autoantibodies to extracellular matrix protein in serum.

BXO occurs in all age groups, from infants to the elderly, most commonly in uncircumcised men. According to Fekete's study, BXO was more common in 51-60-year-old men, with 49,3 % having diabetes and 55,4 % having higher BMI.  $^{1,2}$  A previous study mentioned that most people with BXO seek medical help  $\pm 6$  months after first onset and are already in stage III of the disease. The cause of delayed diagnosis could be most patients present to medical consultations if sexual intercourse is affected, a visible or painful lesion appears, and underdiagnosis by the physician due to a lack of knowledge and a diagnostic indication of the disease. Psychological problems due to embarrassment, shame and emotional stress could also lead to delayed diagnosis. In our case, the patient decided to come due to voiding retention after having the symptoms for years. This caused delayed treatment and management. BXO is rarely treated in the primary care setting.

The clinical presentation of BXO can be summarised as the 6S mnemonic: school age, secondary phimosis, severe sclerotic sclerogenous glandular lesions, and meatal stenosis. The most typical symptoms are the inability to retract the prepuce, discomfort after micturition, and obstructive signs. Secondary phimosis is also common. The voiding dysfunction was rare but caused a high morbidity. The



Fig. 4. Catheter inserted post-meatoplasty.

most common complications are secondary to progressive sclerosis, painful erection, and altered flow. Meatal and urethral involvement in BXO may cause significant disorders and can lead to renal failure.  $^3$  It is estimated that 2–8% of cases developed premalignant change or invasive squamous cell carcinoma.  $^{6,7}$ 

BXO management aims to reduce and relieve symptoms, scars, and atrophy. Pharmacological treatment includes topical steroids and immunomodulators. Mometasone furoate 0,05 % and clobetasol propionate 0.05 % are effective, particularly in mild to moderate BXO, but ineffective in severe BXO. Intralesional triamcinolone injected at the end of preputioplasty has also been used as an alternate treatment. In paediatric cases, circumcision is the definitive management of BXO. In this case, the patient experienced complications with meatal stenosis, so we conducted meatotomy and meatoplasty as required. Topical corticosteroids should continue postoperatively as surgery is not always curative. Lowenstein and Zeichner used intralesional adalimumab to achieve mild BXO in the case of refractory BXO. G. Collective data from the UK in 2000 showed 107 patients (20 %) with BXO urethral stricture; during long-term follow-up, the stricture recurrence rate was almost 90 %. A newer study published in 2016 stated that the recurrence rate

meatal BXO after surgery reached 40 % after a median of two years. 11

The follow-up should be based on the clinical appearance and the patient's complaint. We should ask the patient to come at least monthly for the first 6 months and then can be elongated to visit every 3 months. Whenever the complaint occurs, we should treat it as urethral stricture management. Examination using uroflowmetry, urethrocystography and even following Direct Visual Internal Urethrotomy could performed based on the finding. Treatment of post-op infection and long-term surveillance for recurrence or development of epithelial neoplasia are recommended measures. A scarce complication of lichen sclerosis is squamous cell carcinoma, verrucous carcinoma, and Queyrat erythroplasia, corresponding to a 9.3 % malignancy rate.

#### 4. Conclusion

In summary, balanitis xerotica obliterans is an underreported and undertreated disease. Early diagnosis and immediate treatment are necessary, as it could lead to a urology emergency. Long-term follow-up for recurrence rate and the development of epithelial neoplasia is recommended.

#### CRediT authorship contribution statement

**Rizky Lukman Hakim:** Writing – review & editing, Supervision, Data curation, Conceptualization. **Aninda Dinar Widiantari:** Writing – original draft, Data curation, Conceptualization.

#### **Declaration of competing interest**

The authors declare that they have no competing interests.

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