

## Original Article

# The surgical treatment of Balanitis Xerotica Obliterans

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### ABSTRACT

**Background:** Balanitis Xerotica Obliterans (BXO) is a chronic, often progressive disease, which can lead to phimosis and urethral stenosis, affecting both urinary and sexual function. Steroid creams are usually the first-line treatment but have a limited role and surgical intervention is frequently necessary. Conservative surgical procedures (circumcision) are often preferred in the first instance with the premise that recurrence of disease will require a more definitive reconstruction. This study looked at patients with pathologically proven BXO referred to the Plastic Surgery Unit at James Cook University Hospital between 2005 and 2009. The aim was to look at their management in the past and subsequent management by us. We also looked at whether early referral of progressive and recurrent BXO patients to reconstructive surgery could have prevented unnecessary delay in resolving symptoms at an earlier stage. **Materials and Methods:** Data was collected retrospectively and information regarding the exact anatomical location affected, the extent of the disease, the referring specialty and any previous surgical interventions was obtained. Alterations in urinary and sexual function and relief of symptoms following reconstructive surgery were analysed. **Results:** Of the 23 patients in the study, 43% had previous surgery and 60% of those had undergone two or more procedures. Twenty-one percent of patients had a history of BXO for over five years. Forty-seven percent of patients had alteration in their urinary function and 48% alteration in their sexual function due to the disease, prior to referral. Early results showed remarkable improvement in urinary and sexual function following reconstructive surgery in this group. **Conclusions:** Steroid creams have been shown to limit the progression of the disease but do not offer a cure in the majority of cases. Circumcision can be a curative procedure in early disease. Although there is conflicting evidence for treatment of recurring urethral strictures, repeated urethrotomy or urethral dilatation has poor long-term outcome. In patients with recurrent disease and associated complications we propose early referral to a plastic surgeon with genitourinary interest or reconstructive urologist for definitive treatment.

### KEY WORDS

Balanitis Xerotica Obliterans; penile lichen sclerosis et atrophicus; surgical treatment of Balanitis Xerotica Obliterans

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### INTRODUCTION

Balanitis Xerotica Obliterans (BXO) is a common penile disease, first described in 1928 by Stuhmer.<sup>[1]</sup> It has been classified as a male variant of lichen sclerosis<sup>[2]</sup> by the International Society for the Study of Vulvovaginal disease and is a chronic inflammatory process. Whilst the

exact aetiology is still poorly understood it is thought that it could be genetically determined and there is evidence to suggest a higher incidence of autoimmune diseases amongst patients with BXO.<sup>[1,3]</sup>

The disease usually presents initially on the glans penis or prepuce,<sup>[4]</sup> affecting the foreskin, meatus and distal urethra either individually or in combination. Left untreated it has been reported to affect the entire urethra, penile skin and scrotum.<sup>[1,5]</sup> BXO is most commonly found in patients aged 30-60 years<sup>[4]</sup> but there have been increasing reports of the disease in the paediatric population and it is now known to be the most common cause of pathological phimosis in boys.<sup>[6]</sup>

The clinical progression of BXO can vary; it can present acutely with erythema and discharge leading to early blistering and fissuring or it can follow a more chronic course, presenting initially with grey-white skin discolouration leading to complications at a much later stage.<sup>[1]</sup> In either case, patients can experience ulceration of the glans, fissuring, phimosis, meatal stenosis and urethral strictures in untreated disease leading to problems with urinary and sexual function. On many occasions, progressive disease may lead to long urethral strictures. There have been cases reported of scrotal fistula formation,<sup>[7]</sup> renal impairment<sup>[7]</sup> and a documented association between chronic BXO and an increased risk of penile squamous cell carcinoma.<sup>[8]</sup>

The diagnosis of BXO clinically is almost unmistakable. The differential diagnoses include lichen planus, scleroderma, leukoplakia, vitiligo and erythroplasia of Queyrat. Preoperative diagnostic biopsy is recommended. The histological features are characteristic, showing hyperkeratosis with atrophic epidermis, thinned rete pegs, vacuolar degeneration of basal layer, "Washed out" appearance of the papillary and reticular dermis, amorphous band by dermal collagen at the dermal-epithelial junction and chronic inflammatory infiltrate deep to the band.

Treatment options can be divided into medical or surgical. Topical steroid creams are the mainstay of medical treatment used in early-stage disease but there is limited evidence to support their use in recurrent, severe or advanced disease. Surgical intervention will depend on the disease progression. Circumcision can be a curative procedure for disease confined to the foreskins or glans. But for patients with BXO involving the meatus or urethra the options include meatoplasty, urethrotomy, urethral dilatation or a more definitive procedure such as urethroplasty using

buccal or bladder mucosal grafts and or excision of BXO with skin grafting if involving the glans, coronal sulcus and adjacent shaft of penis. In our unit the senior authors have an established hypospadias practice including adult salvage surgery and additionally offer male genital reconstruction for patients with advanced BXO. We present a review of the literature and report our outcomes following reconstructive surgery.

## MATERIALS AND METHODS

All patients with BXO referred to the Plastic Surgery Department at our unit (tertiary referral centre) between 2005 and 2009 were included in the study. Data was collected retrospectively using a data collection sheet to collate information from patient notes. Referring specialty, the anatomical location of the BXO and duration of symptoms were noted. Previous management including surgical procedures was documented and any change in urinary or sexual function secondary to the disease process was recorded. This was compared to function following plastic surgery management.

## RESULTS

Twenty-three patients aged between four and 78 (mean age 38) [Figure 1] were referred from six different specialties. Forty-eight percent of referrals were from Urology, 17% were from General Practitioners (GPs) and 13% were from Dermatology. The remaining patients were referred from the General Surgery, Genitourinary Medicine and other Plastic Surgery units.

Forty-seven percent of patients were seen at our unit within two years of being diagnosed with BXO but 21% of patients were not referred for over five years and two patients were

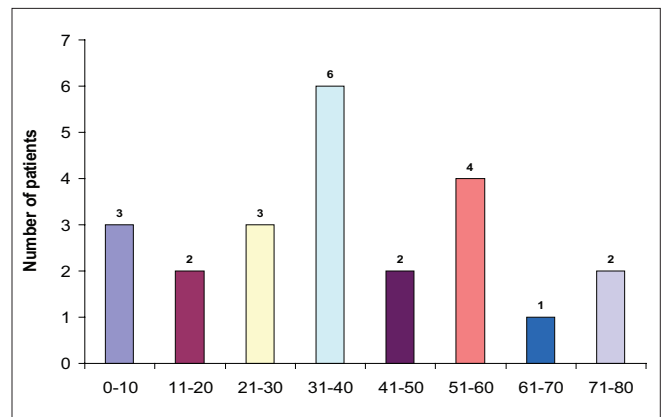


Figure 1: Age group

suffering from BXO-related problems for over ten years.

In the group of 23 there were seven patients who had BXO affecting between three and five areas of their genitalia, indicating severe progressive disease. Of the four that had had previous surgery prior to referral to our unit, all had either release of adhesions or urethral/meatal dilatation. Reconstructive surgery with excision of the affected area and skin grafting or urethroplasty was performed for these patients and they are still being followed in the clinic. To date there is no recurrence of the disease and the patients have reported subjective improvement in their urinary and sexual function from before surgery. A patient in our study who had BXO affecting his meatus and urethra had undergone two optical urethrotomies and four urethral dilatations over a period of 12 years [Figure 2]. Due to the spread of the disease he needed to perform intermittent self-catheterisation twice a week to maintain a reasonable urinary flow for nearly ten years. We performed distal urethrotomy of the dense stricture to allow urethroscopy which also showed skip lesions of BXO in the posterior urethra. The patient underwent excision of the distal stricture and two-stage urethral reconstruction using a buccal mucosa graft [Figure 3]. Subsequently he was able to pass urine normally without the need for the self-catheterisation. At two years post procedure he remains disease-free.

In order to assess the progression of the disease, we recorded the anatomical location of the affected area. Patients were recorded as having BXO affecting the foreskin, glans, meatus, penile shaft, urethra and scrotum or any combination of

these. Forty percent of patients had BXO confined to the foreskin and glans and a further 14% had disease affecting the foreskin only. Thirteen percent of patients had disease affecting three areas, a further 13% had disease affecting four areas and one patient had BXO involving the foreskin, glans, meatus, urethra and scrotum.

Forty-three percent of patients had surgical intervention prior to referral and 60% of those had two or more procedures. These included optical urethrotomy, circumcision, urethral dilatation, hypospadias repair, meatoplasty and debridement/release of penile skin [Figure 4]. Thirty percent had previous medical treatment in the form of steroid creams, anti-fungal cream, antibiotics or self-catheterisation. Sixty-one percent of patients had previously undergone circumcision (a proportion of these were unrelated to the diagnosis of BXO).

Forty-seven percent of patients had alteration of their urinary function due to the disease. Symptoms included reduced flow, hesitancy and pain. Forty-eight percent had problems with sexual function and described fissuring, painful intercourse, bleeding, erectile dysfunction and discharge.

Fifty-two percent of the procedures performed in our department were examination under anaesthesia (EUA) and circumcision [Figure 5]. This correlates with the proportion of patients diagnosed with BXO affecting only the glans or foreskin.

However, four patients underwent a two-stage urethroplasty with buccal mucosal reconstruction, two patients had urethral tubularisations and one had an optical urethrotomy. The remainder had excision of the affected skin and grafting [an example shown in Figure 6] or meatal reconstruction. Early results showed that 100% of these patients had a subjective improvement in urinary or sexual function following plastic surgery input. One hundred percent patients had no recurrence of the disease after treatment in our unit although 13% had BXO still visible but not advancing. Patients were followed up for an average of two years and five months prior to discharge.

## DISCUSSION

When looking at the effectiveness of medical therapy for the treatment of BXO our results show limited success. Many cases eventually required surgical treatment. Of the seven patients treated with steroid creams, anti-fungal

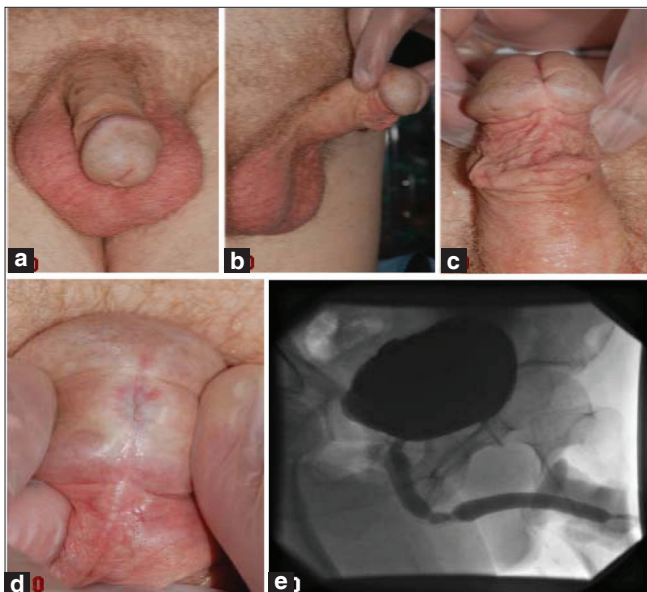


Figure 2: Chronic BXO urethral stricture Pre-op (a-d) and cystourethrogram (e)

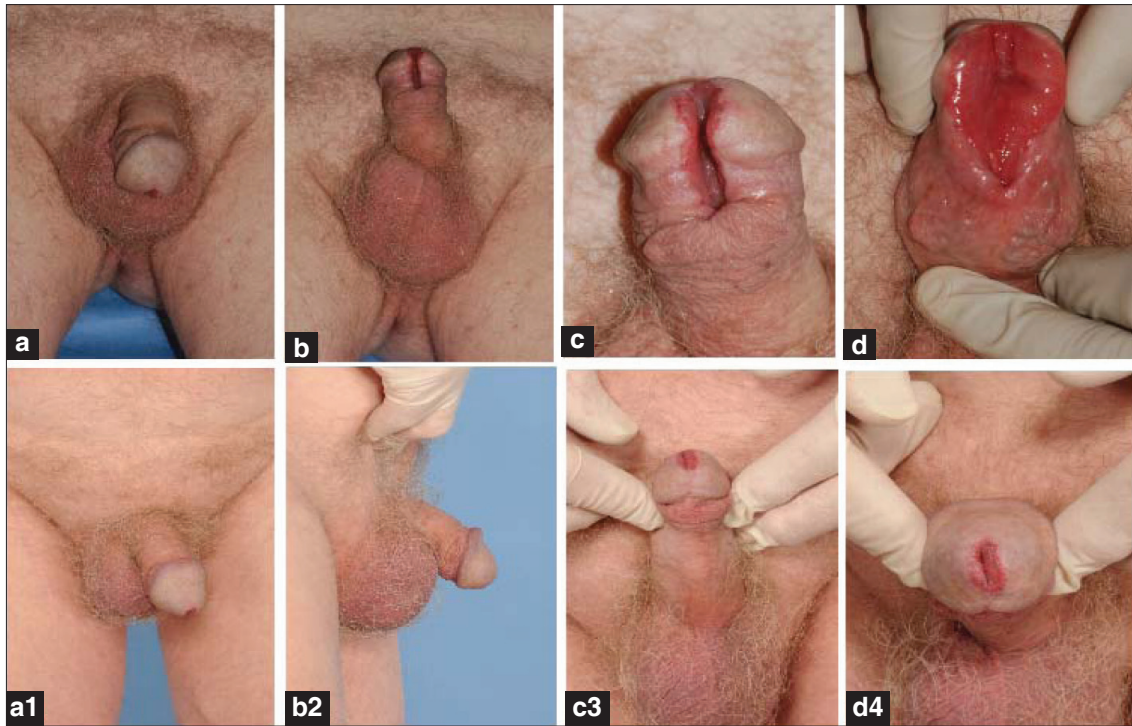


Figure 3: Post-op I stage -stricture excised (a-d) grafted using buccal mucosa & II stage reconstruction (a1-d4)

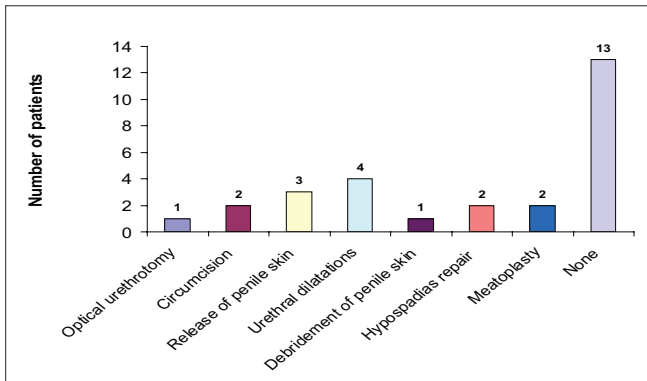


Figure 4: Previous surgical treatment

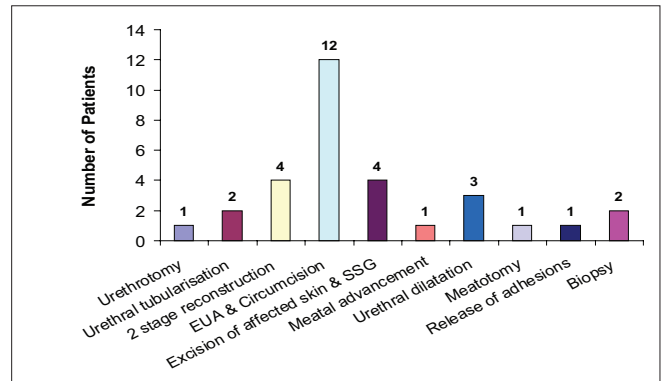


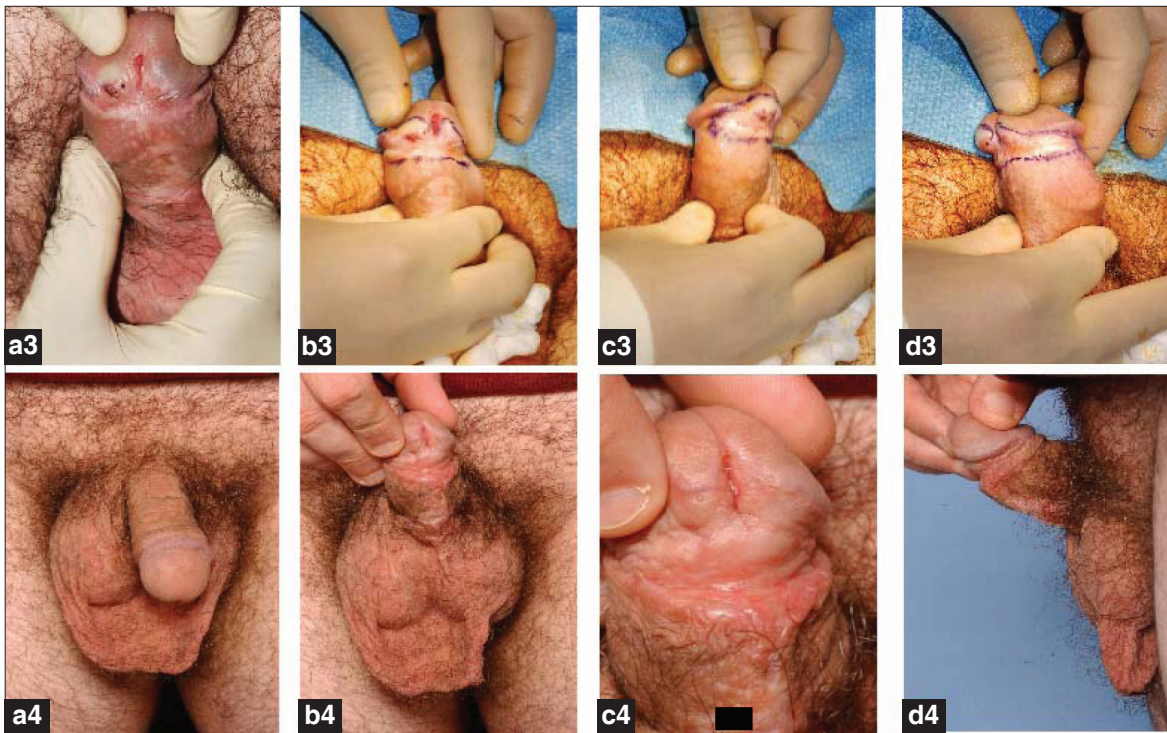
Figure 5: Surgical treatment

or antibiotics three had disease confined to the foreskin and glans but with evidence of adhesions or scarring. The remaining four patients had advanced disease that had spread to the penile shaft or scrotum. A literature review conducted by Pugliese *et al.*, in 2008 concluded that there was no guidance on the duration of treatment with a steroid cream or the type of steroid to use<sup>[9]</sup> and it described limited long-term benefit and disease recurrence once treatment ceased. This was supported by a study looking at a paediatric population using steroid cream which showed resolution in patients with clinically mild BXO that was limited to the prepuce but no reported benefit in those with more severe disease.<sup>[1]</sup> Another paediatric study has shown a recurrence of symptoms following cessation of the steroid treatment

despite initial disease regression.<sup>[10]</sup>

For patients with disease confined to their glans or foreskin, circumcision offers cure by eliminating the moist urine-rich environment in which BXO can progress. In a case series of 287 patients with BXO limited to the foreskin or glans treated by circumcision, 92% had cessation of symptoms and no further advancement of the disease following surgery.<sup>[1]</sup> Our results support this finding and these patients, who had minimal urinary and sexual function symptoms prior to surgery, were discharged from clinic at an early stage.

For patients with urethral or meatal involvement surgical intervention is required. In the case of meatal stenosis



**Figure 6:** BXO involving glans and corona (a3-d3) excised and resurfaced with split skin graft (a4-d4)

or navicular fossa strictures, ventral meatotomy, dorsal meatoplasty or excisional meatoplasty have all been reported.<sup>[9]</sup> Urethral strictures have a high associated morbidity, causing fibrosis of the corpora spongiosum and urinary outflow obstruction, which in turn can lead to high pressures in the upper urinary tract.<sup>[9]</sup> The current gold standard treatment is urethroplasty.<sup>[11]</sup> This method requires more specialised surgical training and a longer hospital stay than conventional methods such as urethral dilatation or urethrotomy. For these reasons, simple procedures are often chosen in preference to urethroplasty but they have limited success rates

Urethral dilatation has never been considered to be curative but is a simple procedure that can be offered under a local anaesthetic in a day case setting. In 1997, Steenkamp *et al.*,<sup>[11]</sup> performed a study directly comparing urethral dilatation to urethrotomy for the treatment of urethral strictures. No statistical difference was found between these two procedures in terms of stricture recurrence rate but both procedures had poor outcomes if used for the treatment of strictures over 4 cm in length. Primary urethroplasty was suggested in such cases.

A recent Cochrane review looked at the evidence comparing urethral dilatation, urethrotomy and urethroplasty for the treatment of urethral strictures.<sup>[12]</sup> Whilst there was

not enough current evidence to compare dilatation and urethrotomy to urethroplasty, there was no difference found between dilatation and urethrotomy in terms of cure rate. A study directly comparing urethral dilatation and urethrotomy found there was no long-term benefit in a second procedure (either dilatation or urethrotomy) for early stricture recurrence.<sup>[13]</sup> If urethroplasty is chosen as a treatment method, options for graft donor site include local genital skin or buccal or lingual mucosa. Trivedi *et al.*, had limited success with local grafts and favoured lingual over buccal mucosa due to easier graft harvesting and lower rates of donor site morbidity.<sup>[14]</sup> Pugliese *et al.*, report high rates of disease recurrence in genital skin grafts and advocate the use of buccal grafts in either a one-stage or a two-stage reconstruction.<sup>[9]</sup>

Depasquale *et al.*, reported 90% stricture recurrence rate in a long-term follow-up of 42 cases of BXO urethral strictures excised and reconstructed using skin, many requiring reoperations using mucosa subsequently. There were no recurrences reported in their series of BXO urethral reconstructions carried out using buccal or bladder mucosa.<sup>[1]</sup>

## CONCLUSIONS

Although our case series is small we think our results show the importance of early recognition of unresolved, progressive and

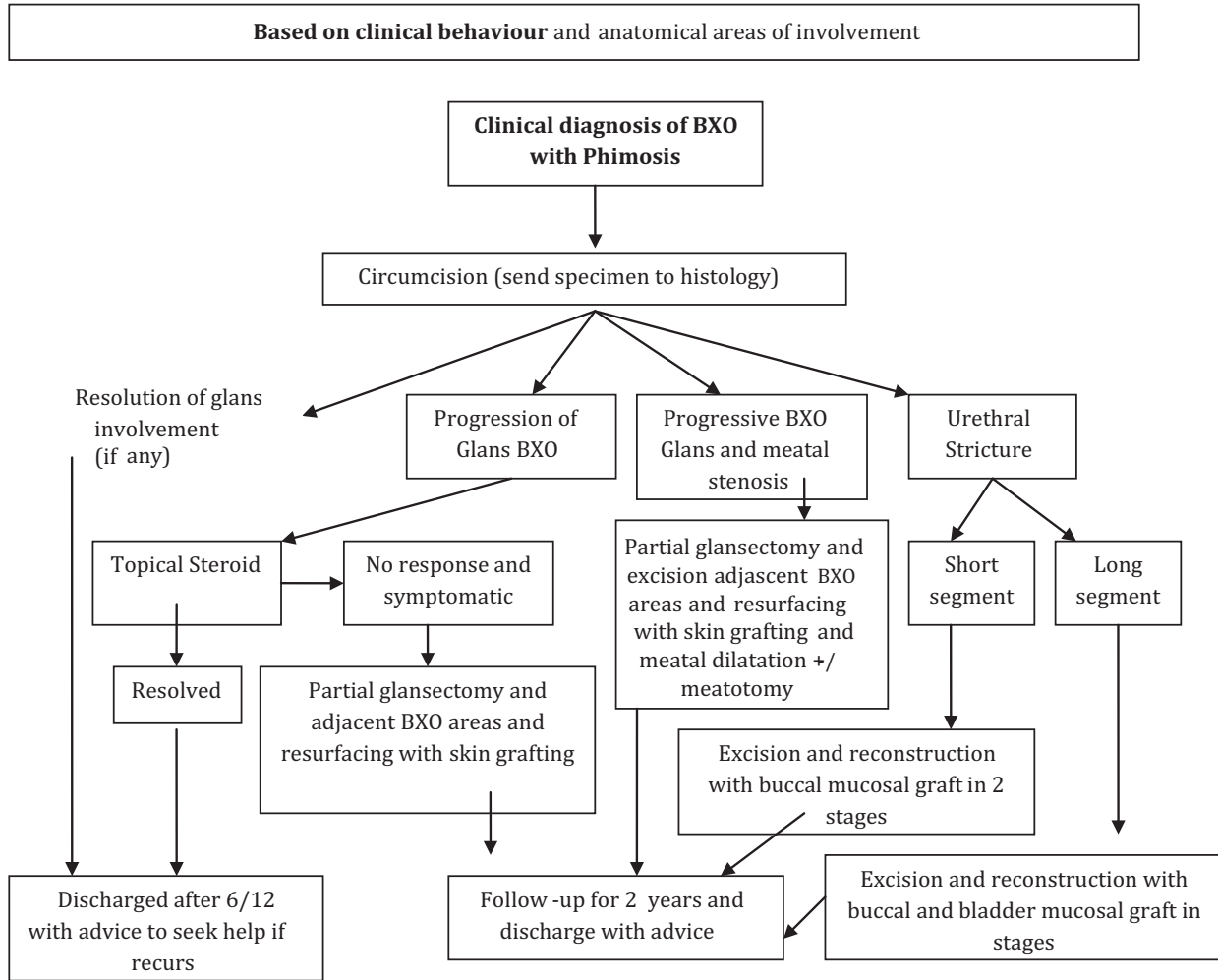


Figure 7: Algorithm for the management of Balanitis Xerotica Obliterans

recurrent BXO cases. Steroid creams have been shown to limit the progression of the disease but do not offer a cure in the majority of cases.<sup>[1]</sup> The progression of the BXO disease gets arrested following circumcision by removing the urine-rich environment and patients with meatal or urethral disease are most likely to require excision and/or reconstruction for a long-lasting cure. Suitable cases should be referred to specialised units for further assessment and surgical management by excision and/or reconstruction for a disease that is prevalent in the male population. An algorithm on the management protocol has been presented [Figure 7].

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