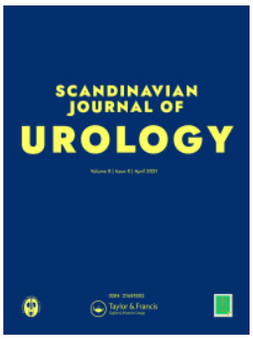

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CASE REPORT

Severe phimosis leading to obstructive uropathy in a boy with lichen sclerosus

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Abstract

Lichen sclerosus (LS) is a chronic, inflammatory condition which commonly involves the anogenital skin. This condition may affect the foreskin, glans, frenulum, meatus and urethra in males. It manifests itself with voiding symptoms. Obstructive uropathy is an extremely rare clinical presentation in childhood. This study reports the case of a 13-year-old boy presenting with obstructive uropathy due to LS and reviews the published data on the diagnosis, management and follow-up of this condition.

Key Words: *circumcision, complication, hydroureteronephrosis, lichen sclerosus, phimosis.*

Introduction

Lichen sclerosus (LC) is a chronic, inflammatory condition which commonly involves the anogenital skin [1]. Although the disorder was formerly called balanitis xerotica obliterans, the American Academy of Dermatology recommended using LS instead [2].

LC involves the foreskin, glans, frenulum, meatus and urethra in males. It leads to destructive scarring in the genital area that causes voiding and sexual problems and impairs the quality of life [3]. Phimosis and voiding symptoms are the most common symptoms in boys. Dysuria, obstructive symptoms and a tendency for urinary tract infections may occur owing to sclerotic prepuce, meatal stenosis or urethral stricture [4]. Obstructive uropathy with hydronephrosis secondary to LS is an extremely rare clinical presentation in boys [5]. This study reports the case of a 13-year-old boy with obstructive uropathy and hydronephrosis caused by LS and reviews published data on the diagnosis and management of this condition.

Case report

A 13-year-old boy presented with complaints of dribbling of urine, abdominal pain and swelling in the right scrotum, which worsened over the past 6 months.The patient had been circumcised at the age of 1 year by non-medical personnel. There had been no significant early complications due to circumcision. However, he had had several fever attacks and had been given antibiotics by the primary care physicians, but had undergone no further evaluations during past 5 years.

The physical examination revealed greyish white discoloration on the sclerotic foreskin and scrotum. Glans penis was concealed due to severe phimosis. The skin of the penis and scrotum was strained. Therefore, at first glance, it was look like a partial

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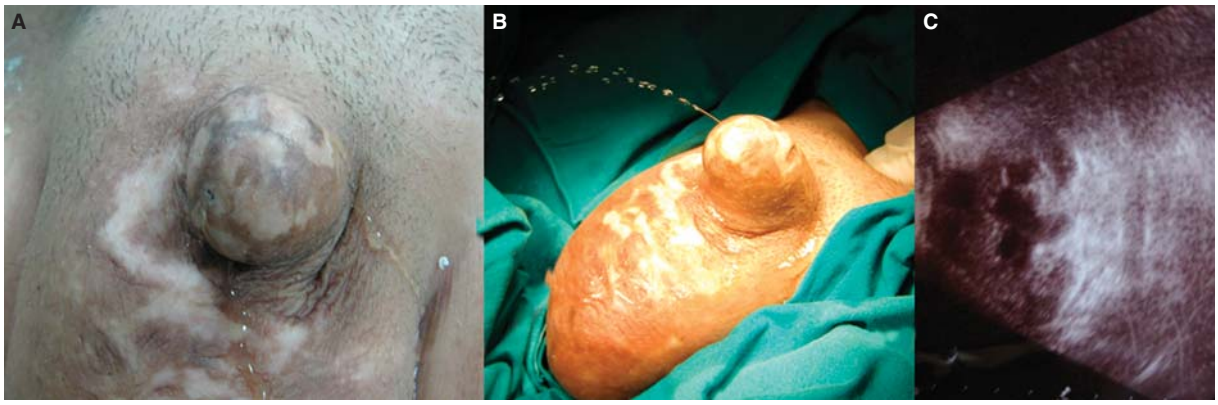

Figure 1. (A) Greyish white discoloration on the sclerotic foreskin and scrotum. Glans penis was concealed owing to severe phimosis. There were two pinpoint openings located ventrally and dorsally. (B)The patient was able to void by applying suprapubic pressure. A giant inguinal hernia sac was palpable in the scrotum (C) Severe to moderate hydroureteronephrosis was detected bilaterally by ultrasonography.

buried penis. There were two pinpoint openings located ventrally and dorsally. A giant inguinal hernia sac was palpable in the scrotum (Figure 1). Ultrasonography revealed globe vesicale, moderate to severe bilateral hydroureteronephrosisand right inguinal hernia (Figure 1C). The biochemical parameters were in the normal range.

At surgery, the tip of a pair of forceps was passed gently along the posterior orifice to ensure the absence of any adhesion between the glans and scarred foreskin (Figure 2A). The cicatrix tissue was incised over the forceps. A large amount of smegma was observed filing the space between the foreskin and glans (Figure 2B). After scarred foreskin resection, circumcision was performed (Figure 3A). The glans penis and meatus were normal. A 12 Fr Foley catheter could easily be inserted. Therefore, no further endoscopic examination to rule out any urethral stricture was performed. The right inguinal hernia was repaired successfully.

Histopathological examination of the resected foreskin revealed LS. The epidermis showed orthokeratotic

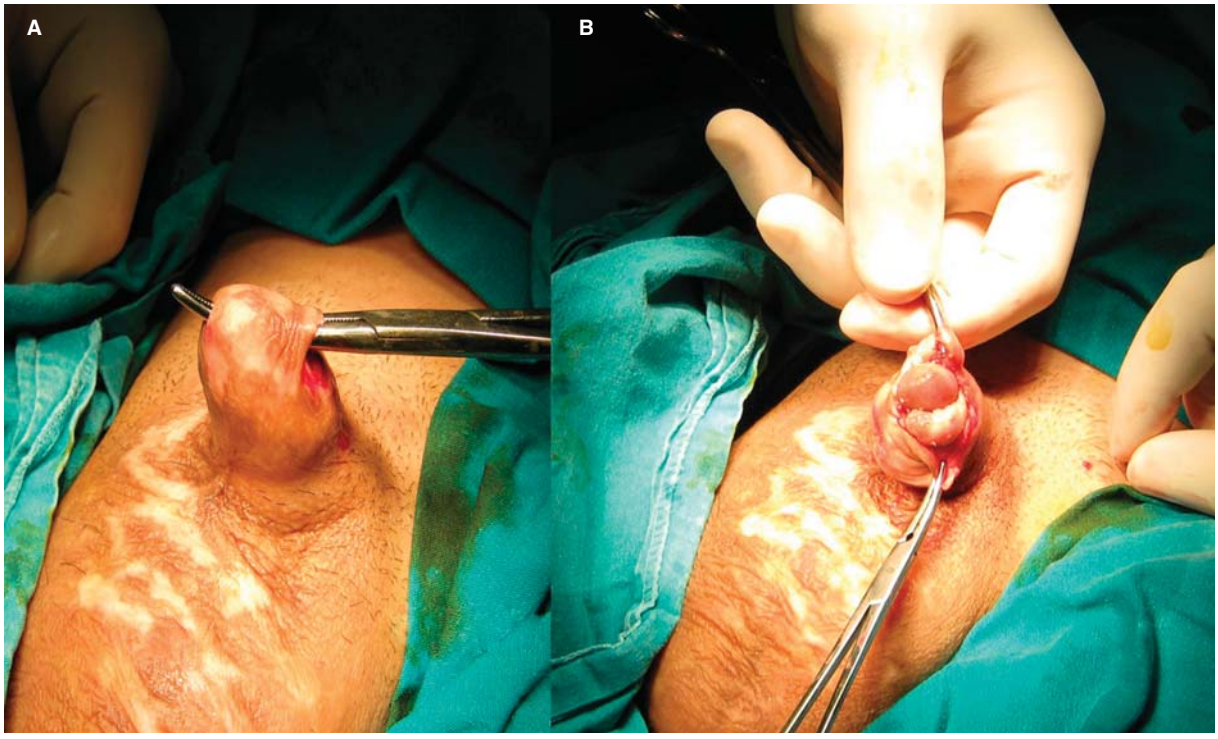

Figure 2. (A) The tip of a pair of forceps was passed gently along the posterior orifice to ensure the absence of adhesions between the glans and cicatrix tissue. (B) A large amount of smegma filled the space between the cicatrix tissue and glans.

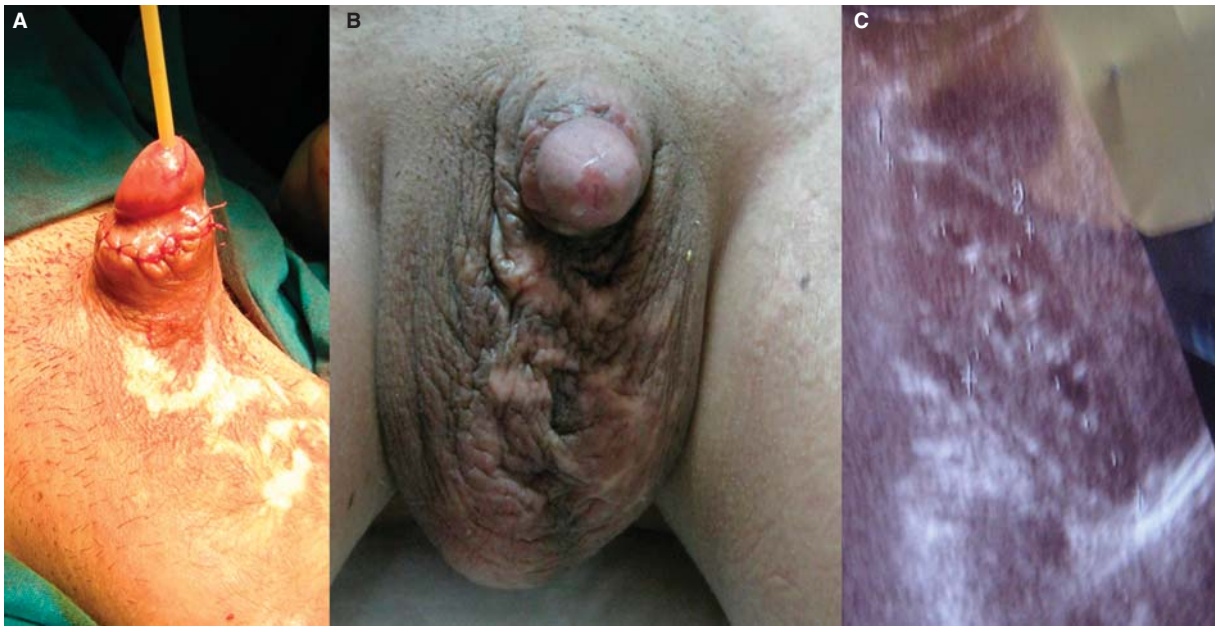

Figure 3. (A) After the removal of the scar, circumcision was performed. (B) The penis was normal 1 week postoperatively. (C) The hydroureteronephrosis resolved spontaneously 4 weeks after the operation.

hyperkeratosis,vacuolar degeneration of the basal layer andsubepidermal oedema. The dominant feature was of broad condensation of the dermal collagen. The Foley catheter was removed at the firstweekcontrol (Figure 3B). The urine calibration was normal with no postvoiding residue. The hydroureteronephrosis resolved spontaneously 4 weeks after the operation (Figure 3C). The parents of the patient were informed about the course of the disease and risk of malignant transformation and requirement for long-term follow-up.

Discussion

LS is a chronic, inflammatory condition of the skin leading to depigmentation and progressive destructive scarring [6]. The disease is more prevalent in women than in men. However it occurs much earlier in males [7]. Although the exact aetiology remains unknown, although immune dysregulation, genetic factors, several infective agents, hormonal influences and local factors are proposed as the precipitating factors [6]. In the present case no causative agent could be detected.

The true incidence of the disease is unknown because of insufficient recognition andthe lack of pathological assessment of the tissue removed at circumcision [1,8]. A prospective studyfound that 40% of the 1178 boys presenting with phimosis had evidence of LS [8]. They concluded that the true incidence of disease is higher than previously assumed in boys with phimosis. Although the exact diagnosis is made by histopathological examination, Meuli et al. described "6Ss" to aid in the diagnosis of LS: school age, secondary phimosis, stenosis of the meatus, severe, sclerotic and sclerogenous lesions of the glans [9]. The present case was of school age and had severe phimosis.

The most common clinical presentation of anogenital LS in boys is phimosis. In a retrospective review, it is reported that it was limited to the foreskin and glans in 57%, meatus in 4% and urethra in 20% of cases [10]. Therefore, difficulty retracting the foreskin is the most common presenting symptom. Pruritis, soreness and obstructive symptoms such as dysuria and poor urinary stream are other frequent complaints. In addition to these symptoms the penis was concealed owing to severe phimosis in the present case. The presence of a right giant inguinal hernia was contributing to penile embedding.

Christman et al. recently reviewed the literature regarding urinary tract obstruction and renal failure associated with LS [5]. They found only six cases of obstructive complications secondary to LS in boys. LS with concomitant hydroureteronephrosis and renal failure was reported in two cases. In these patients the disease was limited to the foreskin and meatus. Although there was bilateral hydroureteronephrosis, biochemical tests were fortunately in the normal range in the present case, and the hydronephrosis resolved spontaneously at follow-up.

Pugliese at al. suggested an algorithm for the medical and surgical management of LS [1]. There

is no cure for LS, but treatment offers control of the condition. They are three reasons for treating LS: relief of symptoms and discomfort; prevention of any or further anatomical changes; and a theoretical prevention of malignant transformation. Although many treatments have been suggested to treat LS over the years, only potent or ultrapotent corticosteroids remain the treatment of choice. After initial therapy, some patients may use corticosteroids only as needed, while others may requiretwice-weekly maintenance therapy. There is no place for surgery in uncomplicated LS. Surgery should be limited exclusively to patients with malignancy and to correct scarring secondary to the disease. Therefore, circumcision is a sufficient surgical procedure in most cases that have severe phimosis due to LS. Appropriate circumcision has very high cure rates in patients with LS limited to the foreskin and glans [9,10]. However, recurrence is reported in the presence of inadequate skin excision [10]. In the present case all scar tissue was excised and sent for pathological evaluation.

The Kobner phenomenon describes the development of typical lesions after injury to uninvolved skin of patients with certain dermatological diseases [11]. Thus, it is essential that patients who have LS after circumcision are followed up for many years.

In conclusion, because of the high incidence of LS in boys with phimosis, all tissue removed at circumcision should undergo pathological examination. Appropriate circumcision remains a sufficient and curative treatment modality for the patients with LS presenting with phimosis. Urethral and meatal stricture should be ruled out in the presence of obstructive symptoms. Patients with LS require long-term follow-up because of the risk of malignant transformation.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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