

Advanced Lichen Sclerosus Resulting in Complete Labial Fusion and Urinary Retention in a Postmenopausal Patient

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Received: 12 July 2018; Accepted: 01 August 2018

Citation: Erich T. Wyckoff, David S. Schweer, Girard Cua. Resident Perception of Obstetrical Hospitalists as Parturient Care Providers and Educators. *Gynecol Reprod Health*. 2018; 2(4): 1-3.

ABSTRACT

Lichen sclerosus is a benign, chronic dermatological condition of unknown etiology that primarily affects the genitalia. The mainstay of therapy is a topical high-potency corticosteroid. Although benign, it is progressive and, in advanced stage disease, can lead to severe functional impairment requiring surgical management. We present a case of advanced lichen sclerosus that resulted in complete labial fusion and obstructive urinary retention requiring surgical intervention. We also performed a review of the literature on similar lichen sclerosus cases. A 71-year-old patient was referred to gynecologic surgery from their urologist for evaluation of complete labial fusion with associated urinary retention. A vulvoperineoplasty was successfully performed. Postoperatively, she was started on estradiol cream daily to assist with initial wound healing. After 19 days, the vagina remained patent and was healing properly. Four months later, she was successfully titrated to twice weekly dosing of clobetasol without recurrence of symptoms. In addition to successful surgical intervention, this case illustrates the need for frequent follow-up and maintenance therapy to prevent relapse.

Keywords

Gynecologic surgical procedures, Lichen sclerosus, Urinary retention, Vagina, Wound healing.

Introduction

Lichen sclerosus (LS) is a benign, chronic, and progressive condition in which inflammation and dermal changes of the anogenital region results in thickening and thinning of the skin. It mostly affects women in the prepubertal, perimenopausal, and postmenopausal periods. The characteristic lesions appear as white, atrophic papules that coalesce into plaques. They can also appear as hemorrhagic, bullous, and eroded ulcers and can undergo secondary lichenification from scratching and the Koebner phenomenon. Though the etiology is unknown, several mechanisms have been proposed, including genetic factors, immune abnormalities, hormonal factors, cell kinetics, and local factors [1]. The disease is usually treated with high-potency topical steroids, which are effective in treating the disease if identified early enough. Severe or advanced disease, however, can lead to functional limitations and even disfigurement. This complicates

the management because surgical intervention may be required, which ranges from lysis of adhesions to vulvoperineoplasty [1].

Here, we describe a case in which a female patient presented at our clinic after being referred by the urologist for long-standing urinary obstructive symptoms in which cystoscopy could not successfully be performed. An exam revealed significant labial and introital fusion consistent with severe LS. The patient ultimately underwent vulvoperineoplasty in addition to topical steroid treatment with good results. As part of this case presentation, we performed a literature search for similar LS cases requiring operative procedures and briefly reviewed surgical technique and suggestions for optimal postoperative management for these types of patients.

Case Report

A 71-year-old patient (gravida 2 and para 2) was referred to our office from the urologist for evaluation of complete labial fusion with associated urinary retention. The patient agreed to participate in this case report and signed a consent form.

Prior to being seen in our office, the patient had a long-standing history of obstructive urinary retention for the past 9 months. An attempt was made by the urologist to perform cystoscopy, but she had significant labial fusion with only a small pinpoint opening through which the cystoscope could not pass. She was prescribed estradiol vaginal cream and referred to our office for further evaluation. She denied having a prior vulvar biopsy or history of LS. Upon presentation, she was complaining of pelvic pain radiating into her lower thighs bilaterally and difficulty voiding. Her physical findings were remarkable for complete labial fusion (Figure 1A).

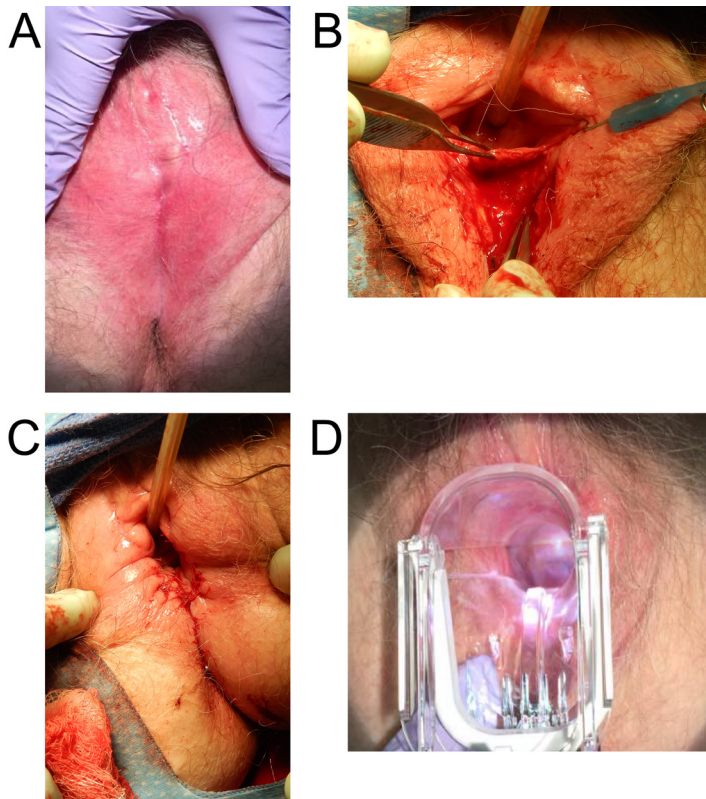


Figure 1: (A) Complete labial fusion during pre-surgery. (B) The creation of the vaginal flap. (C) The translocation of vaginal epithelium over the excised perineum. (D) The 1-year follow-up demonstrating a patent vagina and resolution of urinary retention.

The labial skin was noted to have a cigarette paper appearance with erythematous margins. She was sent from our office to the emergency room for immediate treatment of her urinary retention. While there, a Foley catheter could not be placed because the urethral meatus could not be visualised. The patient managed to produce some urine on her own, and her pelvic pain improved. She was discharged home afterwards. Nineteen days later, she had another bout of urinary retention in which a Foley catheter was again unable to be placed during her visit to the emergency room. She was urgently evaluated by an urologist and a suprapubic catheter was inserted. Approximately 800 mL of urine was drained from the bladder. She was later seen in our office, at which time vulvar biopsies were obtained and found to be indicative of LS with no evidence of malignancy. The suprapubic catheter

allowed for the ability to proceed with either medical or surgical management of her advanced LS. She elected to proceed with medical management and was started on 0.05% clobetasol once daily with plans for re-evaluation at 6 weeks and at 12 weeks after starting therapy. After completion of 12 weeks of medical management, there was no significant anatomical improvement. The decision was made to proceed with surgery. Clobetasol was discontinued 16 days prior to surgery, and the patient underwent vulvoperineoplasty with prophylactic transvaginal Foley catheter placement (Figure 1B-C). The suprapubic catheter was removed by the urologist. Postoperatively, she was started on estradiol cream daily to assist with initial wound healing. Nineteen days later, the vagina remained patent and was healing properly. The Foley catheter was removed, estradiol cream was discontinued, and clobetasol therapy resumed daily for 1 week and then every other day for 3 weeks. Four months later, she was successfully titrated to twice weekly dosing of clobetasol without recurrence of symptoms. She eventually transitioned to a clobetasol maintenance dose of once weekly without recurrence of symptoms a year after surgery (Figure 1D).

Discussion

This case is worthy of mention because of the rarity of complete labial fusion resulting in urinary retention from LS in an adult patient and the severe circumstances of the patient's presentation that required urgent care and a comprehensive review consisting of medical and surgical treatment options. In preparing this report, we performed a literature search for instances in which LS caused labial fusion and were able to find a handful of similar cases.

Ziglioli et al. [2] reported on a 68-year-old patient who presented with urinary tract infections and stress incontinence. She was found to have recurrent LS resulting in labial fusion. Interestingly, she had undergone vulvoperineoplasty 12 years prior and then was pharmacologically treated with topical clobetasol. She was lost to follow-up and presented at that time with labial re-fusion. She underwent repeat vulvoperineoplasty with reportedly good results [2]. This case highlighted the importance of close follow-up with maintenance therapy and the possibility of repeat surgery being successful should LS recur to a similar extent in the same patient.

LS usually occurs in women between 40 and 60 years of age, but can also be seen in the pediatric population, even in children less than 2 years of age [1]. Marcus-Braun et al. [3] reports on a 17-year-old patient who presented with a one-day history of acute urinary retention. She had no prior symptoms, but there was fusion of the labia minora from the clitoris to the vaginal fourchette with a white band of adhesion upon exam. She was taken to the operating room and her adhesion was manually separated by grasping each labium gently. Biopsies were taken and returned positive for LS. She was prescribed clobetasol with close follow-up [3]. In addition, Gibbon et al. [4] described a case series in which patients aged 4 and 3 years presented with labial rash and recurrent urinary tract infection, respectively. Both were found to have near-total labial fusion upon exam. Biopsies later revealed LS in both patients. In these cases, topical steroids were sufficient to resolve the fusion, and a trial of

super potent steroids for 3 to 6 weeks was recommended in this patient population prior to considering surgical treatments [4].

In the adult population and in cases where surgical intervention is necessary, our technique for vulvoperineoplasty is as follows: The urethra is palpated and a lacrimal dilator is used to penetrate the vaginal cavity at the midline inferior to the urinary meatus with minimal risk to the surrounding tissues. Once entrance into the vaginal cavity is confirmed, the opening is gradually dilated and subsequently transitioned to labialplasty. The fused labia are separated superiorly and inferiorly using blunt and sharp dissection. Next, vulvoperineoplasty is performed by creating a transverse incision with the scalpel at the level of the posterior fourchette. The posterior vaginal wall is dissected to create a flap (Figure 1B), and a u-shaped patch of perineal skin and fibrous perineal body is identified and excised so it can be covered by the vaginal epithelium. Next, the vaginal flap is drawn down and sutured in place, covering the denuded perineum (Figure 1C). Since vaginal tissue is rarely affected by LS, part of the posterior vaginal wall is used in the repair to prevent recurrent adhesions and fissuring at the introitus.

Release or excision of adhesions without vaginal advancement is seldom successful [1]. Ziglioli et al. [2] describes a similar surgical technique, though in less depth. Often times the separation of the labia via sharp and blunt dissection alone will not result in long-term relief and patients are at risk of recurrence and re-fusion if not managed well postoperatively. Because of this, we routinely prescribe a maintenance regimen of topical 0.05% clobetasol [1]. Furthermore, adherence to a long-term maintenance therapy to maintain normal skin color and texture reduces the risk of scarring and is associated with reduced risk of differentiated vulvar intraepithelial neoplasia and vulvar cancer [5].

Advanced LS can have detrimental effects on quality of life.

To help ensure long-term relief of the patient's symptoms, we recommend the following key components of patient care:

- Topical 0.05% clobetasol as first line medical therapy.
- Submission of biopsied vulvar and perineal tissue to pathology to further rule out malignancy.
- Surgically, the creation of a vaginal flap to be displaced over the denuded perineum in advanced adult cases refractory to medical management alone.
- Prophylactic postsurgical placement of a Foley catheter to assist with maintaining vaginal patency in the immediate postoperative period and to reduce the risk of urinary retention recurrence.
- Use of estradiol cream in postmenopausal patients postoperatively to aid in wound healing.
- Establishing a maintenance dose of 0.05% clobetasol and close outpatient follow-up in an effort to discourage relapse.

References

1. Cooper SM, Arnold SJ: Vulvar lichen sclerosus. Barbieri RL, ed. UpToDate. Waltham, MA: UpToDate Inc. <http://www.uptodate.com>
2. Ziglioli F, Fornia S, Ciuffireda M, et al. Lichen sclerosus: a review of literature and a case of an atypical surgical treatment. *Acta Biomed.* 2011; 82: 51-57.
3. Marcus-Braun N, Hasan Z, Szvalb S, et al. Acute urinary retention in an adolescent as the presenting symptom of lichen sclerosus et atrophicus. *J Pediatr Adolesc Gynecol.* 2013; 26: e117-e119.
4. Gibbon KL, Bewley AP, Salisbury JA. Labial fusion in children: a presenting feature of genital lichen sclerosus? *Pediatr Dermatol.* 1999; 16: 388-391.
5. Lee A, Bradford J, Fischer G. Long-term Management of Adult Vulvar Lichen Sclerosus: A Prospective Cohort Study of 507 Women. *JAMA Dermatol.* 2015; 151: 1061-1067.