

A Rare Case of Recurrent Labial Fusion Secondary to Lichen Planus Presenting with Voiding Dysfunction

Liken Planusa Sekonder Gelişen ve İşeme Disfonksiyonu ile Ortaya Çıkan Nadir Bir Tekrarlayan Labial Füzyon Olgusu

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ABSTRACT Lichen planus is a T-cell mediated chronic inflammatory mucocutaneous disease of unknown cause. Patients with vulvar lichen planus present most often with itching, burning, post-coital bleeding, dyspareunia, and dysuria. The term 'urinary tract infection' reflects an infection of the urinary system causing an inflammatory response. A threshold of 3 urinary tract infections in 12 months is used to signify recurrence. In this report we introduce a sixty-year-old woman who had been diagnosed with vulvovaginal lichen planus 15 years ago in another center and had been treated with topical steroids. To us, the necessity of her sixth operation in order to separate her recurrent labial fusion which is the cause of her voiding dysfunction and recurrent urinary tract infections, makes this case unique. In conclusion we want to state that further studies investigating the optimum therapeutic regimens and establishing certain algorithms are required for the management of vulvovaginal lichen planus patients.

Key Words: Lichen planus; recurrence; urinary tract infections

ÖZET Liken planus sebebi bilinmeyen T-hücre aracılı kronik inflamatuvar mukokütanöz bir hastalıktır. Vulvar liken planusu olan hastalar genellikle kaşıntı, yanma, postkoital kanama, dispareuni ve dizüri ile başvurumaktadırlar. İdrar yolu enfeksiyonu terimi üriner sistemde inflamasyona yol açan enfeksiyonu temsilen kullanılmaktadır. 12 ayda geçirilen 3 enfeksiyon sonrasında tekrarlamaya özelliği kazanmasından söz edilir. Bu olgu sunumunda 15 yıl önce bir diğer merkezde vulvovajinal liken planus tanısı alan ve topikal steroidlerle tedavi edilen 60 yaşında bir hastayı bildirdik. Tekrarlayan idrar yolu enfeksiyonuna yol açan tekrarlayan labial füzyonu açmak için altıncı kez opere edilmesi, bize göre oldukça nadir bir vaka olduğunu ortaya koymaktadır. Sonuç olarak, vulvovajinal liken planuslu hastaların yönetiminde optimum terapi rejimleri ve kesin algoritmaları ortaya koyacak çalışmaların yapılma gerekliliğini bildirmek istedik.

Anahtar Kelimeler: Liken planus; nüks; üriner kanal enfeksiyonları

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Partial or complete adherence of labia minora and/or majora is defined as labial fusion. A variety of reasons causing vulvovaginal inflammation or irritation can lead to this condition. Although it is relatively common in children, rarely seen in adults and elderly.¹

Lichen planus (LP) is a T-cell mediated chronic inflammatory mucocutaneous disease of unknown cause.² It may involve, separately or simultaneously, different muco-cutaneous sites (oral and genital mucosa, skin, scalp and nail).³ Vulvovaginal LP is characterized by violaceous or erythe-

matous papules or annular plaques or erosions with or without a lacy white border.⁴ These lesions may ulcerate. If only vulvovaginal involvement is present, then disease is more likely to be erosive, with most lesions around labia minora, clitoris, clitoral hood and vagina.⁵ Patients with vulvovaginal lichen planus present most often with itching, burning, pain, postcoital bleeding and dyspareunia.² Progression to scarring of vulva, labial fusion, resorption of labia minora, vaginal adhesions, vaginal obliteration secondary to desquamative vaginitis and increased risk of squamous cell carcinoma are potentially severe complications of the disease.^{6,7} These complications often lead to sexual dysfunction and urinary problems such as voiding dysfunction, dysuria and recurrent urinary tract infections (UTIs).

In this case report, we present a patient with a long standing history of vulvovaginal LP and recurrent labial-vaginal fusion causing voiding dysfunction and recurrent UTIs which are uncommon complications.

CASE REPORT

A sixty-year-old woman applied to our tertiary center's outpatient unit with the complaint of voiding difficulty, dysuria, history of frequent UTIs and adhesion of her labia minora and majora. In her past medical history, she delivered two times vaginally, had hypertension and was postmenopausal for the last 12 years. She had been diagnosed with vulvovaginal lichen planus 15 years ago in another center after a vulvar biopsy which had been performed due to pain, itching and reddish-bluish lesions on her external genitalia. She had been treated with topical steroids after the diagnosis, but her lesions had not regressed despite this therapy and a progressive labial fusion and vaginal obliteration had ensued leading to voiding difficulty and recurrent UTIs. Then, she had undergone her first labial separation surgery. She also mentioned that, during these 15 years, she had been examined by many gynecologists and dermatologists and had additional four operations due to the recurrence of her labial fusion. After these operations only a temporary improvement had been achieved, labial fu-

sion and urinary tract complaints had recurred in a short time. The last operation was 1 year ago in another center and two months later her labial fusion was recurred despite topical steroid and estrogen treatment.

On admission to our hospital, the patient suffered from voiding difficulty with a very thin urinary stream, postvoiding urine dripping and urgency symptoms. Postvoiding residual urine was 330 cc. Our local examination revealed fused labia majora and minora at the midline extending from posterior fourchette to clitoris, covering urethral meatus and vaginal walls and also a reddish well-demarcated area where the urine passes out (Figure 1). A speculum could not be applied. General physical and dermatological examination was normal other than external genitalia. Urinalysis revealed pyuria and the urine culture was positive (100 000 *Escherichia coli*/mL).

Three days after the initiation of antibiotic therapy, under general anesthesia, these labial adhesions were separated manually by simple traction and 3 cm of vaginal walls were dissected bluntly (Figure 2). External urethral meatus seemed normal. Deeper dissection in the vagina was not carried out because this region was densely adhered and the patient was sexually inactive due to severe vulvar pain caused by local inflammation, irritation and adhesions. So it was postponed after the treatment of inflammatory condition.



FIGURE 1: Fused external genitalia.

(See for colored form <http://jinekoloji.turkiyeklinikleri.com/>)



FIGURE 2: Postoperative view of external genitalia.
(See for colored form <http://jinekoloji.turkiyeklinikleri.com/>)

She was discharged at the same day and topical clobetasol 17-propionate 0.05% cream, nitrofurazone 0.2% pomade and topical estrogen cream was prescribed. She was recommended to apply a rolled gauze soaked with topical steroid and antibiotic to the interlabial sulcus to maintain her labia in a separate position just after the application of topical creams for prevention of recurrence. On her control examinations at first and fourth postoperative weeks, the patient reported significant improvement on her urinary symptoms, the surgical site was free of adhesions, but erythematous and chronic inflammatory appearance of the vulva persisted and this area was very tender on palpation. Same topical therapy was recommended to the patient lowering the dosage of estrogen and a literature search was performed for treatment of the nonhealing inflammatory condition. But, later the patient was lost to follow-up.

DISCUSSION

Patients with vulvar lichen planus present most often with itching, burning, postcoital bleeding, dyspareunia and pain.² These symptoms prevent the patient from coital activity. Continuing local inflammation with the absence of coital activity leads to labial adhesion. Early diagnosis of lichen planus has vital importance for not only to treat the patient, but to protect her social life as well.⁸ Development of labial fusion may decrease the sever-

ity of these symptoms but might cause voiding dysfunction and recurrent urinary tract infections which are the chief complaints of our patient.

The possible reasons for labial fusion may be summarized as estrogen deficiency occurring in post-menopausal women, decreased or absent sexual activity, factors related to non-specific and specific inflammatory skin conditions (lichen sclerosus et atrophicus, vulvar lichen planus, genital herpes), poor local hygiene, local trauma, vaginal laceration following childbirth, female genital cutting (infibulation), and recurrent urinary tract infections.^{1,9,10}

Lichen planus is a chronic, pruriginous inflammatory dermatosis, affecting skin, mucous membranes, scalp, and nails. It has polymorphous features. The pathogenesis of lichen planus is not certain, though many studies have supported an immunologic mechanism. The role of T lymphocytes has been emphasized with cytotoxic activity of the CD8+ subset of T lymphocytes responsible for keratinocyte damage.¹¹⁻¹³ And also another major topic is the role of psychological factors in LP. To the results of previous studies, when compared with controls, LP patients have higher stress and anxiety levels and are more prone to depression.¹⁴

There are three clinical variants of LP affecting the vulva: erosive, papulosquamous, and hypertrophic. The most common variant of this disease is erosive lichen planus.¹⁵ This severe form is characterized by violaceous erosions that look like glassy, reticulated, white papules and plaques and, if allowed to progress, can lead to extensive erosion and ulceration with destruction of the vulvar architecture.¹⁶

There are no good evidence based treatment recommendations for the vulvovaginal lichen planus. Only some case reports are present describing the efficacy of oral corticosteroids, azathioprine, griseofulvin, retinoids, dapsone, hydroxychloroquin, topical cyclosporin and topical tacrolimus (FK506).¹⁷

Kirtschig et al. studied 44 patients with mucosal vulvar lichen planus. All women were treated

with potent to very potent topical corticosteroids; however, in the majority of patients symptoms persisted. Only in seven (16%) patients vulvar lichen planus was in remission after a disease duration between 2 and 18 years (mean 10.6 years). These women were left with variable degrees of mucosal atrophy (narrow vaginal introitus, atrophic/fused labia minora) but had no more inflammatory lesions. One patient developed a vulvar squamous cell carcinoma. They reported two vaginal fusion cases in their series.¹⁷

The presented case is encountered very rarely in clinical practice, because there was not only labial and vaginal fusion due to lichen planus but also the resulting voiding dysfunction and the sixth operation was undertaken to treat the condition.

For prevention of labial fusion recurrence after surgery; suturation of the mucosal margins, postoperative topical antibiotics in combination with topical estrogens or steroids, use of vaginal tampons, careful personal or assisted hygiene are suggested.^{9,10,18} But, there are very few data in the literature to prevent the recurrence of labial fusion in intractable cases. Stany et al. reported a patient with

recurrent LP presenting with erosive thinning of the vulvar and vaginal mucosa, loss of the labia minora, and complete vaginal agglutination. The patient failed both medical and conservative surgical management, then underwent a skinning vulvectomy and extraperitoneal simple vaginectomy with a successful neovagina surgery with acellular dermal graft. By 9-month follow up, she continued to do well with return of sexual function.^{18,19} In the presented case, initially a more radical operation could have been considered as in Stany's report, but we preferred a conservative surgical management firstly.

We decided to report this rare case because there are no evidence based therapeutic regimens which are shown to be effectively treating vulvovaginal LP lesions. Moreover, there are no effective surgical methods to prevent the recurrences of labial fusion after surgery other than some radical approaches like skinning vulvectomy and vaginectomy. Further studies investigating the optimum therapeutic regimens and establishing certain algorithms are required for the management of vulvovaginal LP patients.

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