ODÜ Tıp Dergisi / *ODU Journal of Medicine*
http://otd.odu.edu.tr

Olgu Sunumu	***Case Report***
Odu Tıp Derg	*Odu J Med*
(2016) 3:34-36	*(2016) 3:34-36*

Acute Urinary Retention Due to Labial Adhesion as the Presenting Symptom of Lichen Sclerosus in an Adolescent Girl

Adölesan bir kızda liken sklerozun prezentasyon semptomu olarak labial adezyona bağlı akut üriner retansiyon

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Yazının geliş tarihi / Received: 7 Nisan 2014/Apr 7, 2014
Düzeltme / Revised: 26 Mayıs 2014/May 26, 2014
Kabul tarihi / Accepted: 20 Haziran 2014/Jun 20, 2014

Abstract

A rare case of acute urinary retention caused by labial adhesion secondary to lichen sclerosus in a sexually inactive adolescent girl is described with emphasis on the unusual presenting symptomatology. An 18 year old virgin girl presented to the gynecological outpatient department of our hospital with acute pelvic pain and bladder distension. Genital examination revealed labia minora fusion from the clitoris to the vaginal fourchette. The labia minora were completely separated by surgical intervention. Skin biopsies revealed lichen sclerosus. Urinary retention may be caused by complete adhesion of the labia minora which is a rare event in postpubertal individuals and is an unusual initial presentation of lichen sclerosus. Lichen sclerosis should be considered in the differential diagnosis of the labial adhesion.

Keywords: Urinary retention, Lichen sclerosus, vulva

Özet

Seksüel olarak inaktif adolesan bir kızda liken skleroza sekonder gelişen labial adezyonun sebep olduğu nadir bir akut üriner retansiyon olgusu tanımlanmış ve liken sklerozun alışılmadık başlangıç semptomatolojisi vurgulanmıştır. On sekiz yaşında virjin hasta akut pelvik ağrı ve üriner retansiyon şikayetiyle hastanemiz jinekoloji polikliniğine başvurdu. Genital muayenede labia minoralarda klitoristen posterior forşete kadar yapışıklık olduğu tespit edildi. Labia minorlar cerrahi müdahale ile tam olarak birbirinden ayrıldı. Cilt biyopsileri sonrası liken skleroz tanısı konuldu. Postpubertal bireylerde nadir görülen bir olay olan üriner retansiyon labia minorların tam yapışıklıkları sebebiyle ortaya çıkabilir ve bu durum liken sklerozun nadir görülen bir başlangıç semptomudur. Liken skleroz labial adezyonların ayırıcı tanısında göz önüne alınmalıdır.

Anahtar kelimeler: İdrar retansiyonu, liken sklerozu, vulva

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Introduction

Labial adhesion is defined as either partial or complete adherence of the labia minora or majora (1). It occurs most often in infants and prepubertal girls, usually associated with low estrogen levels (2). It is uncommonly seen in postmenopausal women related to hypoestrogenic states, local inflammatory and irritative conditions and vulvar dystrophies (3). Labial adhesion after puberty is extremely rare due to abundance of estrogen and it is usually related to surgical trauma, vulvar trauma or sexual abuse, chronic infections and inflammation, female circumcision, dermatological conditions like herpes simplex, caustic vaginitis, and vaginal laceration following childbirth (1,4-7).

Herein we present a case of almost complete labial adhesion with acute urinary retention in an adolescent girl as the presenting symptom of lichen sclerosus.

Case report

An 18 year old virgin woman presented to the gynecological outpatient department of our hospital with acute pelvic pain and bladder distension. She was unable to urinate during the previous 8 hours. She had a history of increasing difficulty of passing urine and pelvic pain during the last menstruation one week before. She had no history of surgical intervention and her medical history was insignificant. Her menstrual cycles were regular since menarche at 11 years of age.

On physical examination, she had normal secondary sexual characteristics. Examination of the genital area showed almost complete adhesion of the labia minora and bladder outlet obstruction (Fig. 1). Pelvic ultrasound and laboratory work up were normal. Upon further questioning she denied any long-term history of vulvar itching or irritation and any prepubertal history of labial adhesions or lichen sclerosus. There was no history of genital trauma, sexual abuse, genital herpes or syphilis.

Dermatological examination of the vulva revealed complete fusion of the labia minora with obliteration of the vaginal introitus and urethral meatus. Marked atrophy and sclerosis in white porcelain color accompanying with telangiectasias were noted in the vulvar area.

After discussion of therapeutic options and informed consent, a small space was created from the weakest point of the adhesion with a thin clamp. She finally performed an exact micturition after the pinhole opening of the introitus. Topical therapy with clobetasol propionate 0.05%, estrogen and 2% clindamycin cream were administrated on the labium minora for three days and the labia minora were manually separated by grasping each labium gently every day. After three days of failed attempts of separation and not obtaining the desired result, the patient was advised to undergo surgical intervention with general anesthesia. The labia minora were completely separated and released from the translucent line of the labial adhesion area. A normal small vagina with intact hymen and normal urethral meatus was seen. Four weeks of topical therapy with estrogen and clobetasol propionate 0.05% and frequent lubrication was followed by a less potent topical steroid treatment postsurgically. Also topical clindamycin 2% cream was administered for an additional one week after surgery as a prophylactic treatment. The postoperative follow-up was uneventful and the patient was discharged on the postoperative 4^{th} day. The punch biopsy of the adhesion area was consistent with the diagnosis of lichen sclerosus. Saline microscopy of vaginal discharge, and culture results were unremarkable. The patient had no complaint on the postoperative 6^{th} month follow-up.

Discussion

Lichen sclerosus refers to a benign, chronic, progressive dermatologic condition characterized by marked inflammation, epithelial thinning, and distinctive dermal changes accompanied by symptoms of pruritus and pain. Lichen sclerosus is 6-10 times more prevalent in women than in men. Vulvar lichen sclerosus can occur at any age but tends to have two peaks of onset: prepubertal girls and perimenopausal or postmenopausal women (8). The true prevalence is not known; the estimated range varries from 1 in 30 elderly women to 1 in 59 women in a general gynecology practice and 1 in 300 to 1 in 1000 patients referred to dermatologists (9).

Lichen sclerosus preferentially affects the anogenital region although any other cutaneous site may also be affected. Of the patients with genital lichen sclerosus, 15-20% have extragenital disease (10).

The cause of lichen sclerosus remains unknown. The patients may present with intractable pruritus or soreness or, more rarely it may be entirely asymptomatic. Characteristic clinical findings are vulvar hypopigmentation and thin, wrinkled atrophic skin in a figure of eight distribution

encircling the vulvar and perianal region. Focal areas of hyperkeratosis, erosions, and fissures are frequently seen. Lichen sclerosus is a scarring disease and therefore some vulvar architectural change is common (10). Burying of the clitoris secondary to midline fusion or labial adhesion may occur. In untreated or severe disease there may be total loss of labia minora.

The major important point is that symptoms in women may be long standing as there is frequently a delay in diagnosis. It is sometimes not recognized and misdiagnosed as thrush or other problems and not correctly diagnosed until the patient is referred to a specialist when the problem does not clear up. Up to 15% of cases are seen in children with the majority starts with vulvar presentations.

In the present case, our patient presented with acute urinary retention for the first time in her life with no other prior complaints. She had no history of chronic vulvar irritation or itching. Thus, lichen sclerosus after puberty is a rare condition and the initial presentation can be unusual. Marcus-Braun et al reported similar case report and noted that, in such cases with acute urinary retention, a suspicion of underlying pathology such as asymptomatic lichen sclerosus should be raised and be confirmed by a biopsy (11).

A high level of suspicion is required in cases of acute labial adhesions in adolescent in order not to misdiagnose lichen sclerosus, a rare disease in this age group. It must be kept in mind that although rare, labial fusion may be the initial presentation in patients with lichen sclerosus and surgical invention may need to be performed in these advanced cases.

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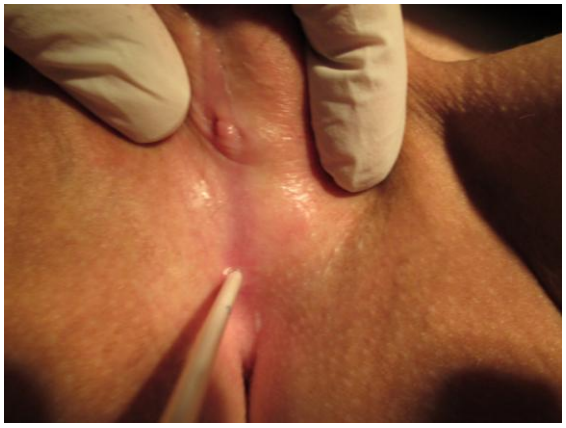

Figure 1. Complete fusion of the labia minora with obliteration of the vaginal introitus and urethral meatus.