Case Report / Olgu Sunumu

Labial Adhesion in Adulthood: A Report of Two Cases

Erişkinde Labial Adezyon: İki Olgu Sunumu

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Abstract

Labial adhesions are generally observed in the premenarchal and postmenopausal period. They are rarely seen in adulthood. Here, two labial fusion cases in the reproductive period are discussed. In the first case, 38-year-old virgin woman presented with anuria to the emergency unit. On her physical examination, labia were fused totally including periurethral area. His history revealed that she was mentally retarded and had primary amenorrhea. She had poor selfcare. On her genital examination, she had secondary sexual characteristics, including axially-pubic hair and breast development at Tanner stage-5. In her surgical treatment, the labia were overturned and a well-defined introitus was constituted. After the operation, topical estrogen prescribed. In second case, a forty-two year old woman with Behçet's disease presented with dysuria and difficulty in voiding for about one month. Her obstetric examination revealed that she had two vaginal births. On pelvic examination, genital ulcer was determined on the labium. Besides, adhesion almost completely fused into the bilateral labia minora. For surgical treatment, manual separation technique was used under general anesthesia. After the operation, topical steroid was prescribed. Labial adhesions are observed, especially in the premenarchal period. However, they may be very rarely seen in the reproductive period due to local inflammation and circulating low estrogen levels. Topical estrogens, topical steroids, oral estrogen treatment, manual separation under anesthesia or without anesthesia, sharp or blunt dissection under anesthesia are alternative treatment methods. The success rates of these treatment methods exceed 90%. Besides, the recurrence rate varies between 4% and 20% with manual and sharp dissection methods.

Keywords
Labial adhesion, adulthood, surgical treatment

Anahtar Kelimeler
Labial adezyon, erişkin dönem, cerrahi tedavi

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Öz

Erişkinlerde nadir görülen ve değişik sebeplere bağlı olabilen labial adezyonlarla özellikle premenarş ve postmenapozal dönemde karşılaşılmaktadır. Burada, farklı nedenle oluşan iki erişkin labial füzyon olgusu takdim edilmiştir. Olgu-1: İdrar yapamama şikayetiyle acil servise başvuran 38 yaşındaki virgo kadının muayenesinde labiumların periüretral alanı da içerecek şekilde tamamen kapattığı görüldü. Anamnezinde, hastanın mental retarde olduğu ve primer amenoresi olduğu öğrenildi. Genel vücut bakımı vasat olan hastanın yapılan muayenesinde meme gelişimi ve pubik kıllanma gibi sekonder seks karakter gelişiminin Tanner Evre- 5 olduğu gözlendi. Olgu 1'in tedavisinde, cerrahi olarak labiumlar dışa devrilerek introital açıklığın devamlılığı sağlandı. İşlem sonrasında östrojenli krem verildi. Olgu-2: Daha önce Behçet hastalığı tanısı alan, yaklaşık bir aydır idrar yaparken zorlanma, yanma yakınmasıyla konsülte edilen 42 yaşındaki hastanın anamnezinde iki normal doğum yaptığı öğrenildi. Hastanın yapılan pelvik

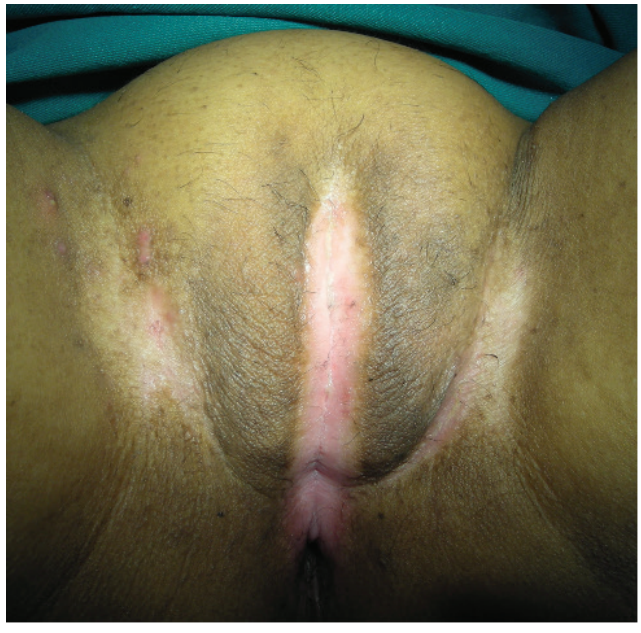
muayenesinde labiumlarda Behçet hastalığı ile ilgili olabilecek genital ülser saptandı. Ayrıca bilateral labia minoralarda totale yakın adezyon saptandı. Olgu 2'nin tedavisinde genel anestezi altında manuel ayırma tekniği uygulandı. İşlem sonrasında topikal steroidli krem verildi. Labial adezyonlar özellikle premenarş döneminde, çok seyrek olarak da reprodüktif dönemde lokal enflamasyon ve düşük östrojen düzeyine bağlı olarak görülebilir. Anestezi altında keskin veya künt diseksiyon, anestezi altında veya anestezisiz manuel ayırma, topikal östrojen, oral östrojen, topikal steroid alternatif tedavi yöntemleridir. Tedavi yöntemlerinin başarı oranı %90'a kadar çıkmaktadır. Rekürrens oranı ise manuel veya keskin diseksiyon yöntemleri ile %4-20 arasında değişmektedir.

Introduction

Labial adhesions usually appear in the prepubertal period (1). Their incidence ranges from 0.6% to 3%. What causes them is uncertain (2). Leung et al. (3) has reported that labial adhesions are not a congenital pathology (3). Chronic irritation, poor hygiene and sanitary napkins in prepubertal hypoestrogenic females can be responsible for their etiology (4). Chronic irritation causes desquamation of the epithelium especially on the labia minor. Thereafter, reepithelization, adhesions and an avascular membrane appear on the labia minor. Formation of labial adhesions is explained by stimulation of inflammation, excessive activation of macrophages and excessive fibrosis in this hypoestrogenic environment (5). Although labial adhesions are relatively common in the prepubertal period, they rarely appear in adults. In this report, two cases of labial adhesions in adulthood are presented. Informed consents were obtained from the patients.

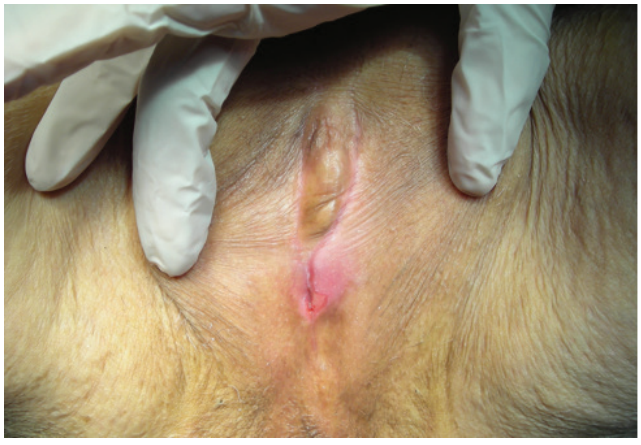
Case 1: A 38-year old virgin woman presenting with anuria to the emergency unit was found to have mental retardation and primary amenorrhea on history. She had poor hygiene habits and fully developed secondary sex characteristics like breast and pubic hair on physical examination. Pelvic examination revealed that the labial adhesion completely covered the vulva including the periurethral region (Picture 1). The woman was found to have difficulty in urination occasionally, which had been relieved thanks to ointments containing estrogen prescribed before. She was treated with surgery; the labia were turned outwards and the introital opening was achieved. An estrogen-containing cream (Estriol cream, 50g, 1x1 application/day, Assos Pharmaceuticals, İstanbul, Turkey) was prescribed. Follow-ups two weeks and forty-five days after her discharge showed that she had sufficient vaginal opening and did not have any problems with urination.

Case 2: A 42-year old woman presented with a two-month history of difficult urination, burning sensation and genital lesion. Diagnosed as Behçet's disease 6-7 years ago, the woman had ocular involvement. Therefore, she was still on treatment with topical (Demovate 0.05% 50 gr cream, 50g, 1x1/day, Glaxo Smith Kline İlaçları Sanayi ve Ticaret AŞ, İstanbul, Turkey) and oral steroids (prednol 16 mg, 1x1/day, Mustafa Nevzat İlaç, İstanbul, Turkey). On History, the woman was found to have two vaginal births. Pelvic examination showed a genital ulcer likely to be due to Behçet's disease. In addition, an adhesion was found to cover the bilateral labia minora almost completely (Picture 2). The patient was treated with manual separation under general anesthesia. A topical steroid-containing cream (Demovate 0.05% 50 gr cream, 50g, 1x1/day, Glaxo Smith Kline İlaçları Sanay, ve Ticaret AŞ, İstanbul, Turkey) was given. The follow-up six months after her discharge revealed that



Picture 1. Labia were fused totally including periurethral area in case 1

the patient had a normal vaginal depth and totally separated labia.



Picture 2. Adhesion almost completely fused into the bilateral labia minora in case 2

Discussion

Labial adhesions appear especially during premenarchal or postmenopausal periods (1). In this report, two cases of labial adhesions emerging in adulthood have been presented. Few cases of labial fusion in adulthood have been reported in the literature. In a series, cases of labial fusion developing in adulthood due to lichen sclerosus or lichen planus and repaired with surgery were described (6). Watanabe et al. (7) reported a case of labial adhesion causing anuria but not creating any sexual problems. In addition, a case of labial adhesion having an opening as small as the eye of a needle and diagnosed in the sixth gestational week was described in the literature (8).

Labial adhesions in adults may have different causes. Sexual abuse can lead to labial adhesions due to chronic irritation and trauma in some cases. The first case presented in this report did not have any signs of sexual abuse or trauma although she had mental retardation. Cases of labial adhesions developing after procedures like vaginal birth have also been reported (9). Among other causes are female circumcision (10) and herpes infections (8). In addition, lack of a long-term sexual relationship leads to recurrent labial adhesions as in postmenopausal women.

Labial adhesion in the first case presented here could be attributed to hypoestrogenism. We wanted to perform genetic tests to determine whether labial adhesion is accompanied by any syndromes in the case having mental retardation and primary amenorrhea. However, the patient's relatives did not accept it due to high costs of the tests. There have not been any cases of labial adhesions accompanied by primary amenorrhea and mental retardation. The second case of labial adhesion presented here had Behçet's disease. Genital ulcers developing in the labia due to Behçet's disease might have caused chronic inflammation and labial fusion.

Most of the cases of labial adhesions in the prepuberty are asymptomatic. Also, hydronephrosis due to difficulty in urination, vulvar irritation, urinary tract infections (11) and even urinary retention in children has rarely been reported in the literature (2). Adults with labial adhesions may complain about an inability to have a sexual intercourse due to dyspareunia and vaginal stenosis (12). It has been stated in the literature that adults have pelvic inflammatory disease due to bacterial colonization of the uterus and uterine tubas caused by urinary retention (13). Both cases reported here presented with difficulty in urination and anuria.

Taking care of vulvar hygiene plays a primary role in the treatment of labial adhesions. It has been reported that using estrogen-containing creams has achieved a rate of success in 50-88% of the symptomatic cases (14). Although there is not an agreement on the duration of treatment, it is recommended that it should last a few weeks (15). The creams containing estrogen can have side-effects such as temporary hyperpigmentation in the labia minora, breast buds, vaginal bleeding and precocious puberty in children. Several studies also revealed recurrence rates varying between 11% and 40% (2). As an alternative treatment, topical steroids applied for 4-6 weeks are very successful (5). Steroids can cause erythema, folliculitis, itchiness, hirsutism and skin atrophy in the short-term (16) and adrenal suppression, growth retardation or cancer in the long-term. In a retrospective study on prepubertal girls, topical steroids were found to separate adhesions more rapidly than estrogen and create lower rates of recurrences. It has been argued that using two topical treatments in combination can be more effective than using them individually, but that no significant differences have been reported (16). In the first case presented here, since we suspected hypoestrogenism was responsible for etiology of labial adhesion,

using topical estrogen treatment twice daily and taking care of vulvar hygiene as much as possible were recommended before surgery. In the second case presented in this report, topical steroids were recommended to suppress the chronic inflammatory reaction. Neither of the cases had side-effects due to these treatments during their follow-up.

Surgery is performed in cases of labial adhesions refractory to topical treatment (2). Thick adhesions are treated under general anesthesia and thinner ones are treated under local anesthesia (4). In some cases of labial adhesions, intranasal midazolam was utilized for surgical separation (1). In addition, amniotic flaps or skin grafts obtained from legs are used for separation in advanced stages of adhesions. Since two cases presented here were resistant to topical treatment, surgical treatment was performed. In the first case, the labia were overturned and the introitus opening was achieved. In the second case, manual separation was utilized. Both cases underwent surgery under general anesthesia and they did not need flaps.

It is still debatable whether estrogen and steroids should be used before surgery. In cases of labial adhesions accompanied by lichen sclerosus or planus, suppression with steroids is recommended before and after surgery (6). Although Rouzier et al. (17) reported that medical suppression is not necessary in the preoperative or postoperative periods, Goldstein and Burrows (18) revealed that postoperative steroid suppression reduces recurrences (8). Tebruegge et al. (19) recommend that estrogen treatment should be used for a few weeks before surgery. They noted that preoperative estrogen treatment improved the surgical outcome. There is not an agreement on the duration of topical treatment (15). We used a six-week topical treatment after surgery in our cases.

In conclusion, labial fusion can appear not only in the premenarchal period but also in adulthood. Depending on its etiology, topical estrogen or steroids can be used in adults. However, surgery is still a successful treatment to eliminate urinary symptoms in cases resistant to topical treatment. While using creams containing estrogen or steroids after surgery or manual separation decreases recurrences of labial adhesions, their effectiveness has not been proven yet.

Ethics

Informed Consent: Informed consents were obtained from the patients.

Peer-review: Externally and internally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: S.N.A., H.Y., Concept: S.N.A., S.D.S., H.Y., Design: S.N.A., S.D.S., H.Y., Data Collection or Processing: S.N.A., H.Y., Analysis or Interpretation: S.Ö.A., H.Y., Literature Search: E.Z., Writing: S.N.A., M.K.

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