CASE REPORT

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Labial Fusion with Urinary Symptoms: A Case Report

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INTRODUCTION

Labial adhesions, also known as synechia vulvae or labial agglutination is a condition characterised by the fusion of the labia minora or majora, and most commonly located near the clitoral area.[1] It can be complete or partial fusion, flimsy or dense, and it may be congenital or acquired. Acquired cases occur mainly in prepubertal girls and postmenopausal women due to oestrogen deficiency.[2] Advanced stages of labial fusion or labial adhesion, although rare, may be diagnosed in postmenopausal and sexually inactive women. In the elderly the incidence is not well elucidated , but the condition accounts for around 0.6% to 1.4% in children.[3] [4] Patients may be asymptomatic or present with urinary or vulval symptoms.[2] When it manifests clinically, the common symptoms are post-void dripping, haematuria, dysuria, and local inflammation in the labial area. We present a case of a postmenopausal lady who presented to our Urogynaecology clinic with a history of dribbling of urine (intermittent incontinence) which progressed to urinary retention.

CASE STUDY

Mrs NC is a 65 year old para- 2 lady. She presented to our Urogynaecological clinic with a 6 month history of voiding dysfunction. She complained of progressively worsening of spontaneous urine dribbling, incontinence and suprapubic discomfort, but no dysuria, nor haematuria. She is a known hypertensive patient with no other co-morbidities. Her surgical history is unremarkable and she has been sexually inactive for more than ten years. She does not smoke nor use alcohol.

On examination she was clinically well with a normal systemic examination. Her initial genito-urinary examination revealed excoriation of the vulva, which appeared flattened but with a bulging mass in the vagina. There was dense fusion (agglutination) of more than 90% the labia majora with only a small opening near the clitoris through which urine was draining (fig.1). A clinical assessment of labial adhesion was made. Her diabetic screen and urine culture was negative. Mrs NC was counselled and offered conservative management with topical oestrogen cream and reviewed after six weeks. The response to the oestrogen cream was poor and an examination under anaesthesia with surgical division of labia was planned. However the patient returned with almost complete fusion of the labia and inability to pass more than some drops of urine before the surgery date. She was subsequently taken to theatre and the adhesions separated with sharp (blade) and blunt (forceps) surgical technique. A biopsy was also taken to exclude malignancy, lichen sclerosus and lichen planus. She is currently discharged to continue with topical oestrogen cream and will be reviewed for histology results, recurrence and long term management and follow-up.

DISCUSSION

According to Norris, the precise etiology of labial fusion remains unknown, but low estrogen states are postulated as a cause. This is evidenced by the rareness of the condition in the reproductive age group where women have sufficient levels of estrogen.[5] The genital area is particularly susceptible to irritation and inflammation in the postmenopausal stage and the risk of adhesion increases in the setting of diabetes mellitus, lichen sclerosis or decreased sexual activity.[1] Risk factors such as low oestrogen levels of menopause, chronic vulvar inflammation, and sexual inactivity in the pathogenesis of labial fusion were reinforced in a study by Kaplan et al.[6] Erickson identified other possible causes such as malignancy, lichen sclerosis, radiotherapy, and topical medications,[7] while Singh reported chronic inflammation due to poor hygiene, eczema, lichen planus or sclerosis, seborrheic dermatitis, eczema, local trauma and recurrent urinary tract infections as possible aggravating factors.[2] Differential diagnoses across age groups include hymenal skin tags, imperforate hymen, introital cysts, Mayer-Rokitansky-Kuster-Hauser syndrome, ureterocoele, urethral prolapse, vaginal atresia and vaginal rhabdomyosarcoma.[1]

There is usually no need for special laboratory investigations nor imaging in the diagnosis of labial adhesion as it is a clinical diagnosis and in asymptomatic patients it may be an incidental finding. A physical examination of the genitourinary system in is imperative in females who present with difficulty voiding, urinary retention or urinary tract infections, prompting early treatment. Gonzalez describes labial adhesion as being commonly located near the clitoral area and consisting of thin fibrotic tissue which could cause either partial or complete fusions that occlude the vaginal orifice, and associated post-void dripping, haematuria, dysuria, and local inflammation in the labial area, which resolve quickly when adhesions resolve.[1]

Conservative management of labial adhesion is a reasonable approach in cases of early or partial labial fusion. This includes the application of topical oestrogen creams, with or without topical steroids. The aim is to block ongoing agglutination of especially labia minora and a restoration of the anatomy. While guidelines are lacking previous reviews recommended two months of conservative treatment before surgical management is considered.[2,8]. The use of betamethasone and clobetasol have been prescribed where Lichen Sclerosis is involved and betamethasone was shown to be superior to topical oestrogen in terms of resolution and side effect profile.[2]

Where there is no response to topical therapy or the presence of complete labial fusion, surgical separation under general anaesthesia will be required.[3,8]

Funda et al. described a technique for surgical management of a postmenopausal women with complete labial fusion using

a combination of cautery (no sutures) and blunt dissection and reported resolution of symptoms and no recurrence at six months follow up. Other surgical management options as reviewed by them include use of Hegar diltators, curved forceps and use of skin flaps to prevent recurrence, all with promising long term outcomes.[3]

Urinary Pseudo-incontinence in postmenopausal women

According to Mikos et al. postmenopausal labial fusion is manifestation of a severe form of menopausal genitourinary syndrome.[8] They reported on a case series of women with an average age of 72, who presented with mainly continuous urinary incontinence and complete labial fusion. All women required surgical intervention which was uneventful with immediate resolution of incontinence and no recurrence of labial agglutination after two years follow up. In a case report of a postmenopausal woman with voiding difficulty and incontinence, Palla reports that she required surgical division of labial adhesions. Surgical management resulted in immediate resolution of her symptoms as confirmed by multichannel urodynamic testing post- operatively.[9] Julia reported on a 72 year old woman with complete labial fusion whose sole presenting complaint was urinary incontinence which resolved after surgical therapy. This was a case of failed conservative treatment with oestrogen cream.[10] Lu et al. reported on an 83 year old woman with labial agglutination, intermittently treated with topical oestrogen over a 3 year period. Surgery instantly resolved her symptoms and an incidental intraepithelial neoplasia was noted. There was no recurrence after 3 months and they emphasised that surgical intervention should be the first consideration for labial agglutination with urinary symptoms in postmenopausal women. Surgery also allows essential evaluation of other high risk conditions (malignancy) in this age group.[11]

Lichen Sclerosus and Lichen Planus

In a study of thirty-five patients with symptomatic labial adhesions due to Lichen Sclerosus (LS) or Lichen Planus (LP), the mean age was 57 years with a mean symptom duration of 9 years. Of the patients, 27 had LS and 8 had LP. Of the 35 patients, 21 had posterior fusion, 11 had anterior fusion, and 3 had both anterior and posterior fusions.[11] MacPherson reports that Lichen sclerosus (LS) and lichen planus (LP) are both immunologically mediated conditions and has a preference for the genitalia. While LP mainly affects mucous membranes such as the mouth and vagina, these are rarely affected by LS.[12] According to Howard et al , vulval lichen planus-lichen sclerosus overlap is an emerging observation. They report on a case of a 63 year old women with a 20-year history of ulcerative vulvo-vaginitis initially diagnosed with cicatricial pemphigoid. A later clinic-pathological diagnosis of LP was changed to LS following further biopsies of lesions in the labia, perineum and peri-anal area. They highlighted the challenge of extensive vulvo-vaginal ulceration.[13] The inflammatory mucocutaneous LS affecting the anogenital areas, affects mostly postmenopausal women and is largely underdiagnosed. Recognised associations include hormonal status, persistent trauma and autoimmune diseases, however infections do not seem to be clear risk factors. LS pathogenesis involves factors such as a Genetic predisposition and an immune-mediated Th1-specific IFNγ-induced phenotype is implicated in its pathogenesis. The clinical presentation, which can be confirmed with skin biopsy, is chronic whitish atrophic patches along with itching and soreness in the vulvar, perianal and penile regions. Genital scarring, and sexual, urinary dysfunction, and squamous cell carcinoma can result from the lesions.[14] While LS can be asymptomatic, it is usually a pruriginous condition. The treatment of lichen sclerosus aims at controlling the symptoms, stopping further scarring and distortion and reducing the risk of cancer. Follow-up must be kept indefinitely.[15]

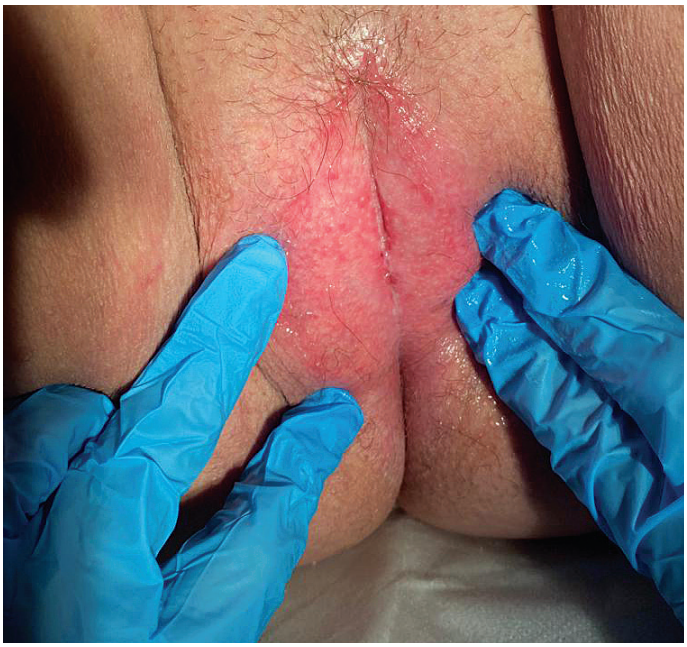
First-line treatment for LS is a super-potent topical corticosteroid ointment which has a high response rate, such as clobetasol propionate 0.05% ointment twice daily. Second-line therapies include topical calcineurin inhibitors (pimecrolimus or tacrolimus) and systemic agents. There is limited evidence for systemic treatments for both conditions. The risk of vulval squamous cell carcinoma (SCC) is increased in both LP and LS, and it is not known how treatment affects this risk.[12] According to Perez-Lopez, surgery which include restoring vulvar anatomy and treating clitoral phimosis, introital stenosis, and vulvar granuloma fissuratum lead to improved sexual dysfunction and satisfactory outcome is used mainly for the treatment of complications associated with lichen sclerosus.[15,16]

According to Krapf, other treatment modalities include platelet rich plasma, high energy modalities (photodynamic therapy, high intensity focused ultrasound, fractional laser therapy), as well as lysis of vulvar lesions and perineoplasty.[16]

CONCLUSION

Labial adhesion, a fusion of the labia minora or majora, is often an incidental finding. The exact cause is unclear but it is believed to occur in a low oestrogen state and diminished sexual activity espescially in postmenopausal women. Aggravating factors include chronic inflammation such as lichen planus or sclerosis, as well as poor hygiene, eczema, seborrheic dermatitis, eczema, local trauma or recurrent urinary tract infections. Complete labial fusion in postmenopausal women can lead to urinary pseudo-incontinence. Treatment consists of oestrogen with or without steroids and surgery in cases of failed conservative treatment or complete labial fusion with urinary symptoms.

Figure 1: Labial Fusion



REFERENCES

1. *Gonzalez D, Anand S, Mendez MD. Labial Adhesions. [Updated 2023 Jan 31]. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2023 Jan*
2. *Singh P, Han HC. Labial adhesions in postmenopausal women: presentation and management. Int Urogynecol J. 2019 Sep;30(9):1429-1432.*
3. *Gungor Ugurlucan F, Yasa C, Tas IS, Aslay I, Yalcin O. Complete labial fusion causing urinary retention in a postmenopausal woman. Am J Clin Exp Urol. 2021 Oct 15;9(5):413-415.*
4. *Laih CY, Huang CP, Chou EC. Labial adhesion in a postmenopausal female: A case report. Medicine (Baltimore). 2020 Jun 26;99(26):e20803. doi: 10.1097/MD.0000000000020803.*
5. *Norris JE, Elder CV, Dunford AM, Rampal D, Cheung C, Grover SR. Spontaneous resolution of labial adhesions in pre-pubertal girls. J Paediatr Child Health. 2018 Jul;54(7):748-753.*
6. *Kaplan F, Alvarez J, Dwyer P. Nonsurgical separation of complete labial fusion using a Hegar dilator in postmenopausal women. Int Urogynecol J Pelvic Floor Dysfunct. 2015;26(2):297–8.*
7. *Eriksen J, Glavind-Kristensen M, Guldberg R. Complete labial fusion in a postmenopausal woman: unusual cause of urinary symptoms. Int Urogynecol J. 2019;30:331–333.*
8. *Bradford J, Fischer G. Surgical division of labial adhesions in vulvar lichen Sclerosus and lichen planus. J Lower Genit Tract Dis. 2013;17(1):48–50.*
9. *Palla L, De Angelis B, Lucarini L, Spallone D, Palla G. Cervelli V. a case of labial fusion and urinary pseudo-incontinence in an elderly woman. A surgical treatment and a review. Eur Rev Med Pharmacol Sci. 2010;14(5):491–3.*
10. *Julia J, Yacoub M, Levy G. Labial fusion causing urinary incontinence in a postmenopausal female: a case report. Int Urogynecol J Pelvic Floor Dysfunct. 2003;14:360–1.*
11. *Lu BJ, Chin HY, Chu CY, Wang JTJ. Postmenopausal labial agglutination mimics urinary incontinence and hidden vaginal lesion. J Obstet Gynaecol Res. 2018;44:801–805.*
12. *McPherson T, Cooper S. Vulval lichen sclerosus and lichen planus. Dermatol Ther. 2010 Sep-Oct;23(5):523-32.*
13. *Howard M, Hall A. Vulval lichen planus-lichen sclerosus overlap. Int J STD AIDS. 2018 Sep;29(10):1017-1023.*
14. *De Luca DA, Papara C, Vorobyev A, Staiger H, Bieber K, Thaçi D, Ludwig RJ. Lichen sclerosus: The 2023 update. Front Med (Lausanne). 2023 Feb 16;10:1106318.*
15. *Pérez-López FR, Vieira-Baptista P. Lichen sclerosus in women: a review. Climacteric. 2017 Aug;20(4):339-347.*
16. *Krapf JM, Mitchell L, Holton MA, Goldstein AT. Vulvar Lichen Sclerosus: Current Perspectives. Int J Womens Health. 2020 Jan 15;12:11-20.*