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Case report

Clinical Manifestations and Treatment of Postmenopausal Labial Adhesion: A Case Series and Literature Review

Nastaran Mahmoudnejad[1*], Farzaneh Sharifiaghdas[1], Shabnam Tehrani[2], Mohammad Hamidi Madani[1], Arian Karimi Rouzbahani[3], Narjes Saberi[4]

[1]*Urology and Nephrology Research Center, Shahid Labbafinejad Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran*

[2]*Infectious Disease and Tropical Medicine, Shahid Beheshti University of Medical Sciences, Tehran, Iran*

[3]*Student Research Committee, Lorestan University of Medical Sciences, Khorramabad, Iran*

[4]*Isfahan Kidney Disease Research Center, Isfahan University of Medical Sciences, Isfahan, Iran*

HIGHLIGHTS

- Postmenopausal labial adhesion is a very rare entity. Probable underlying causes include: low estrogen levels, poor hygiene, eczema, local trauma, recurrent UTIs, vulvar dystrophies and lack of sexual.
- Unlike prepubertal LA, in adult women surgical treatment is usually necessary for symptomatic and severe cases.
- The increasing number of LA cases highlights the importance of conducting a thorough genital examination in all females with hypoestrogenic state and voiding complaints, especially those who are not sexually active.

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***Corresponding Author**:
Nastaran Mahmoudnejad
Email: nastaran.mahmoudnejad@gmail.com
Address: Labbafinejad Hospital, 9th Boostan Ave., Pasdaran Ave., Postal code:166666311.

ABSTRACT

Introduction

Labial adhesion (LA) is rarely observed in postmenopausal women. There is no uniform consensus regarding the etiology of the LA, yet. It may present with different urinary or vaginal symptoms. We herein report the clinical symptoms, management and follow-up of eight postmenopausal patients with LA and a brief review of the literature.

Case presentation

We reported the presenting complaints, treatment and surgical outcomes of eight postmenopausal patients, diagnosed with LA. The mean age and follow-up were 55 years (range: 42-69) and 27 months (range:18-36), respectively. All patients were either virgins or did not have sexual intercourse for years. The increasing number of adults LA case reports highlights the importance of conducting a thorough genital examination in all females with hypoestrogenic state and voiding complaints, especially those who are not sexually active. Placing separate absorbable sutures at the introitus area, prolonged use of topical estrogen and maintaining sexual contact or vaginal cones may help to reduce the early recurrence.

Conclusions

The surgical technique can be considered a safe, effective and durable method for the treatment of LA in postmenopausal women.

Keywords: Female genital, Incontinence; Labial adhesion; Post menopause; Urogenital disease; Vulva

Introduction

Labial adhesion (LA), also known as labial fusion or agglutination, is a disorder characterized by the complete or partial fusion of the labia minora. This condition

typically affects girls before puberty, but it can also rarely be seen in postmenopausal women (1). LA can occur due to congenital anomalies or acquired conditions and is often the result of a combination of estrogen deficiency and chronic inflammation (2, 3). In severe cases of postmenopausal LA, the labia minora completely adhere to each other at the midline, leaving no opening at the introitus. This condition has been associated with pseudo incontinence or vaginal voiding in women (4). LA can be managed simply with topical estrogen, steroids, or manual separation during the prepuberty period. However, there is currently no established standard treatment for postmenopausal women (1). In this article, we review the relevant literature and present the symptoms, treatment, and follow-up course of eight cases of LA in postmenopausal or hypoestrogenic women. To the best of our knowledge, this is the largest case series on LA in the literature.

Case presentation

Case 1

A 69-year-old woman was referred to our medical center complaining of difficult voiding, postvoid dribbling, and straining. These symptoms had first appeared 6 months prior to admission and had been progressively worsening. Her past medical and surgical history was unremarkable, and she had not engaged in sexual intercourse for the past 10 years. Urine analysis and culture results came back normal. An ultrasound study revealed bilateral renal fullness and a postvoid residual volume (PVR) of 400 cc. The patient had undergone multiple conservative and medical treatment courses, but none had provided a cure. During the physical examination, it was observed that the labia minora were fused together, resulting in a complete LA. The urethral meatus was not visible, but a small opening was found in the midline. A hidden clitoris was also noted. As a result, the patient underwent clitoral release and surgical separation of the labia minora (Figures 1a and 1b).

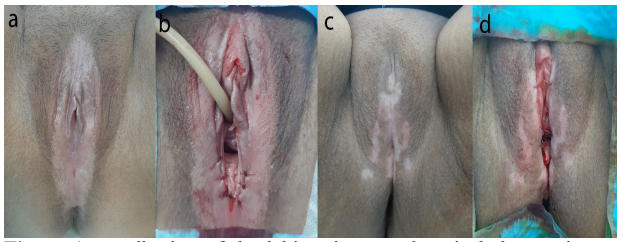

Figure 1. a: adhesion of the labia minora and a pin hole opening at introitus, b: clitoral release and eversion of the introital mucosa with separate absorbable sutures, c: severe LA and skin hypopigmentation due to lichen sclerosus, d: same patient after surgical lysis and clitoral release.

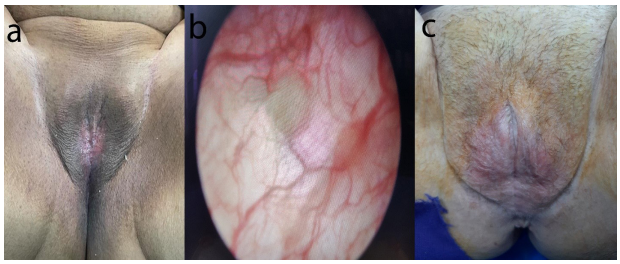

Figure 2. a: severe LA and a pinpoint orifice, b: cystoscopic view of the same patient showing cystitis cystica, c: complete LA

Case 2

A 59-year-old female was referred to urology clinic complaining of frequency and postvoid dribbling. She was a known case of lichen sclerosus. She was sexually inactive for 5 years. During genital examination, we observed complete LA, a pin-point orifice in the posterior part of the fusion and skin discoloration due to lichen sclerosus. Following surgical intervention, prolonged use of topical estrogen and corticosteroids and intermittent use of a vaginal cone, were advised (Figures 1c and 1d).

Case 3

A 68-year-old woman presented with difficult voiding, postvoid dribbling, and recurrent urinary tract infections (UTIs). In an ultrasound study, a postvoid residual volume of 200 cc was reported. She was sexually inactive in the past 7 years. She mentioned a history of abdominal hysterectomy due to abnormal uterine bleeding 15 years ago. On physical examination, a complete LA and a pin-point orifice were seen. The patient underwent surgical labia minora separation. Due to severe urethral stricture, cysto-urethroscopy and urethral dilatation were performed at the same session. In cystoscopic evaluation, multiple translucent submucosal cystic lesions on the posterior wall of the bladder were observed. Pathological evaluation confirmed the diagnosis of cystitis cystica (Figures 2a and 2b)

Case 4

A 57-year-old virgin woman was admitted to the emergency department with urinary retention. She mentioned a history of frequency, urgency, and postvoid dribbling for 2 years. Physical examination revealed LA. Emergent surgical intervention for the separation of adhesions and urethral catheterization was performed successfully (Figure 2c).

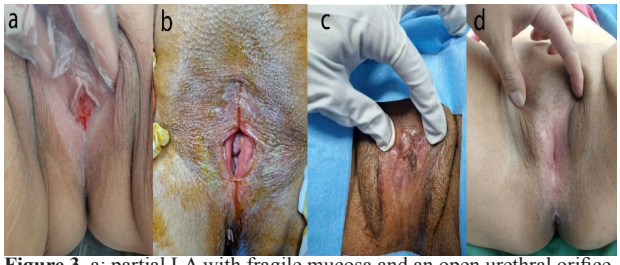

Figure 3. a: partial LA with fragile mucosa and an open urethral orifice, b: partial LA with covered urethral orifice, c: complete LA, d: complete LA with a pinhole orifice

Table 1. Reported cases of LA in postmenopausal women

First Author/ Journal	Publication year	Number of cases	Age (y)	Clinical Presentations	Physical Examination	Underlying condition	Treatment	Follow up
Chuong c./ Obstetrics and gynecology.	1984	1	75	Urinary incontinence, recurrent UTI	Extensive labial fusion	-	Manual separation	-
Savona-Ventura c./Australian and New Zealand Journal of Obstetrics and Gynaecology.	1985	2	78 and 82	vulvar pain, difficult voiding, pseudoincontinence	Labial adhesion	Sexually inactive	Surgical treatment	No recurrence
Imamura R./ Hinyokika Kiyo.	1998	1	68	Dysuria, pseudoincontinence	Extensive labial fusion	-	Separation with the Hegar's dilator	No recurrence
Saito M./Urologia internationalis.	1998	2	78 and 77	difficult voiding vulvitis, urinary retention	Extensive labial fusion	Sexually inactive	Surgical treatment	No recurrence
Ong NC./Australian and New Zealand journal of obstetrics and gynaecology.	1999	1	88	Difficult voiding, postvoid dribbling	Labial fusion	Sexually inactive	Surgical treatment	Some fusion at the 2-month follow-up.
Yano K./Plastic and reconstructive surgery.	2002	1	66	Dysuria, perineal irritation	Severe labial fusion	-	Surgical treatment and Y-V flap	No recurrence at 1-year-follow-up
Hatada Y./Acta Obstetricia et Gynecologica Scandinavica.	2003	1	71	Vulvar pain, abnormal urinary flow	Extensive labial fusion	Sexually inactive	Surgical treatment	No recurrence at 8-month follow-up
Julia J/International Urogynecology Journal.	2003	1	72	Postvoid dribbling, incontinence	Labial fusion	Sexually inactive	Surgical treatment	No symptoms at 2-week follow-up
Pulvino JQ./ International Urogynecology Journal.	2008	5	Mean age= 78	Pseudoincontinence	Complete labial adhesion in 4 patients and partial adhesion in 1 patient	-	Surgical treatment	No recurrence at 2 and 6 week- follow-up
Palla L./Eur Rev Med Pharmacol Sci.	2010	1	71	Pseudoincontinence	Extensive labial fusion	Hysterectomy, cystocele repair	Surgical treatment	-
Dirim A./ International urogynecology journal.	2011	1	73	Recurrent UTI, pseudoincontinence	Fused labia major	Sexually inactive	Surgical treatment	No recurrence at 2- week follow-up
Fakheri T./ The Professional Medical Journal.	2011	1	74	Urinary retention	Total fusion of labia	Sexually inactive	Surgical treatment	No recurrence at 3- month follow-up
Lazarou G./ Female Pelvic Medicine & Reconstructive Surgery.	2013	1	51	Incomplete voiding	Complete labial adhesion	-	Surgical treatment	No recurrence at 3- month follow-up
James R./Open Journal of Obstetrics and Gynecology.	2014	1	79	Pseudoincontinence	Severe labial agglutination	Sexually inactive	Surgical treatment	No recurrence

Kaplan F./ International urogynecology journal.	2014	2	78 and 65	Pseudoincontinence, voiding difficulty, incomplete bladder emptying	Complete labial fusion	Case one: History of Hodgkin's lymphoma, hysterectomy,cervical cancer, melanoma Case two: hysterectomy,lichen sclerosis	Surgical treatment	No recurrence at 1-year follow-up
Prahl R./Emergency Medicine News.	2014	1	85	Abdominal pain, urinary retention	Labial adhesion	-	Medical	-
Başaranoğlu S./ International journal of surgery case reports.	2016	1	92	Acute renal failure	Complete labial fusion	lichen sclerosus	Surgical treatment	No recurrence at 3-month follow-up
Dănău R./ Roman J Urol.	2016	1	71	Obstructive voiding symptoms, urinary pseudoincontinence	Labial fusion	Hysterectomy	Surgical treatment	-
Lu BJ./Journal of Obstetrics and Gynaecology Research.	2017	1	83	Pseudoincontinence	Labial fusion	-	Surgical treatment	Partial labial fusion at 1-year follow-up
Eriksen J./ International Urogynecology Journal	2018	1	82	Obstructive urinary symptoms, stress incontinence	Labial fusion	Sexually inactive	Surgical treatment	-
Kumagai Y./ Journal of Medical Case Reports.	2018	1	76	An elevated accumulation was seen in the vagina on a positron emission-tomography scan	Extensive labial fusion	Chemo-radiotherapy for esophageal cancer	Surgical treatment	No recurrence at 3-month follow-up
Wyman AM./ Obstetrics & Gynecology.	2018	2	90 and 71	Pseudoincontinence, voiding dysfunction	Complete labial fusion	One had lichen sclerosis	Surgical treatment	No recurrence at 12 and 18-month follow-up
Dangal G./ Journal of Nepal Health Research Council.	2019	1	58	Urinary retention	Complete labial fusion	Unmarried	Surgical treatment	-
Takimoto M./ Case Reports Plast Surg Hand Surg.	2019	1	86	Dysuria, perineal pain	Extensive labial fusion	Hysterectomy. Sexually inactive	Surgical treatment , vulvo-perineal flaps	No recurrence at 18-month follow-up
Mikos T./ Case Reports in Women's Health.	2019	7	Mean age= 72.9±12.1	Pseudoincontinence	Complete labial fusion	Sexually inactive	Manual or surgical labial separation	No recurrence at a mean 2.4-year follow-up
Kukreja B./Gyn PAJO.	2019	1	60	Difficult voiding, poor urinary stream	Labial fusion	Hysterectomy- lichen sclerosus	Surgical treatment	The case is on regular follow-up
Singh P./ International Urogynecology Journal.	2019	6	Mean age: 76	Urinary and vulvar complaints	Complete labial fusion	One had lichen sclerosus	4 needed Surgical treatment	No recurrence at 1-month follow-up
Takemaru M./ Case Reports in Medicine.	2019	1	91	Recurrent UTI	Labial fusion	History of two times labial-adhesion separations	Surgical treatment	No recurrence at 6-month follow-up

Laih C-Y./Medicine.	2020	1	76	Voiding difficulty, dribbling, Pseudo-incontinence	Labial fusion	Sexually inactive	Surgical treatment	No recurrence at 6-month follow-up
Saberi N./ Iranian Journal of Medical Sciences.	2020	1	,	Voiding dysfunction and recurrent UTI	Diffuse labial adhesion	Virgin	Surgical treatment	No recurrence at 3-month follow-up
Tanvir T./Journal of Mid-life Health.	2020	1	68	Pseudoincontinence	Complete labial Fusion	Sexually inactive	Surgical treatment	No recurrence at 3-year follow-up
Murugesan1 A./Journal of South Asian Federation of Obstetrics and Gynaecology	2020	1	65	Dribbling and abdominal pain	Adhesion of the labia minora	-	Surgical treatment	No recurrence at 3-month follow-up
Gungor Ugurlucan F./Am J Clin Exp Urol.	2021	1	75	Urinary retention	Complete labial fusion	Sexually inactive	Surgical treatment	No recurrence at 6-month follow-up
Maeda T./ Case Reports in Women's Health.	2021	1	82	UTI	Extensive adhesion of labia majora	-	Surgery (Z-plasty on the ventral side and Y-V-plasty on the anal side)	No recurrence at 8- month follow-up
Williams C./ Urology Case Reports.	2021	1	58	Overactive bladder	Severe labial fusion	-	Manual separation	Multiple episodes of recurrence at 12-week-follow-up

Case 5

A 62-year-old virgin female, who complained of prolonged dysuria, a burning sensation in the genital area, and spotting, was referred to our clinic. Urine analysis revealed microscopic hematuria, and urine cytology results were negative for malignancy. During the physical examination, we detected partial LA with a fragile, easily bleeding mucosa. Despite treatment with topical estrogen for 12 weeks, there was no improvement, so the patient was scheduled for surgical intervention (Figure 3a).

Case 6

A 56-year-old patient presented with postvoid dribbling. In the physical examination, partial LA was observed. Lab tests and imaging studies were all normal. She had not engaged in sexual activity for the past 3 years. Topical treatment including estrogen and steroids proved ineffective, leading to the decision to manage her condition through surgery (Figure 3b)

Case 7

A 55-year-old female complaining of stress urinary incontinence and postvoid dribbling referred to urology clinic. She reported that a significant amount of urine comes out of her vagina immediately after voiding while standing. She was sexually inactive in the past 11 years. In the physical examination, complete LA was noted. She underwent surgical separation of the LA (Figure 3c).

Case 8

A 42-year-old virgin female patient complaining of vulvar pain, dysuria, intermittency, postvoid dribbling and recurrent UTI was referred to the urology clinic. Urine analysis showed urinary infection, and in ultrasound evaluation, a PVR of 150 cc was reported. She mentioned a history of premature ovarian failure in the past 6 years. In the physical examination, complete LA with a hidden clitoris was revealed (Figure 3d).

Surgical technique and follow-up

In two patients diagnosed with partial LA, the first optional medical treatment option was topical estrogen application with simultaneous gentle traction of the labia minora for 3 months. However, neither of them achieved a complete response, so surgical separation of the labial fusion was performed. For all patients with severe and complete LA, the initial treatment modality was surgical intervention under spinal anesthesia in a lithotomy position. The surgical technique was a combination of blunt and sharp dissection starting from the presumed urinary orifice. The adhesions were usually easily released, and cautery was used for hemostasis if

necessary. To prevent early fusion and recurrence, vaginal mucosal eversion in the introitus region was achieved with separate 3-0 absorbable stitches. In cases of hidden clitoris, hoodoplasty or simple release of the clitoris was also considered. Following the surgery, a vaginal tampon and Foley catheter were inserted for one day. All patients were discharged the day after the procedure and advised to take oral antibiotics for 3-5 days. The postoperative course was uneventful. Topical estrogen was prescribed for all patients to be used every day at bedtime for 1 month, every other day for 1 month, and then twice a week for at least one year. Short-term topical corticosteroids, topical emollients, and vaginal cones were also prescribed for selected patients. None of the patients experienced recurrence of LA or any related complications during the 27- month (range: 18-36) follow-up period.

Discussion

LA typically affects prepubertal girls. There are a few case reports on this condition in postmenopausal women (Table 1).The prevalence of this disorder in the elderly is still unclear (1). In infancy, this condition usually occurs as a result of local contamination and irritation caused by diapers. It can also be associated with adrenogenital syndrome and adrenocortical hyperplasia. In the postmenopausal period, LA develops due to low estrogen levels, poor hygiene, eczema, local trauma, recurrent UTIs, and lack of sexual intercourse (2, 3). Vulvar dystrophies, including lichen sclerosus, have also been suggested as underlying condition (3). In our study, all patients were either virgins or had not engaged in sexual activity for a long time.

One patient had a past medical history positive for lichen sclerosus. Vulvar lichen sclerosus is a chronic inflammatory disease that can affect women of all ages and is managed with topical corticosteroids. Due to an increased risk of malignancy, close follow-up is recommended (4). LA can be asymptomatic or present with vaginal symptoms such as pruritus and vulvodynia. In some cases, it may cause voiding symptoms (5). The severity of urinary symptoms may not necessarily match the severity of LA. In early stages, adhesion is seen in the posterior part of the vaginal introitus. However, in severe cases, the vaginal and urethral orifice are entirely covered by adhesion. If urine fails to exit the vagina freely, it can lead to urinary retention, recurrent UTIs or pseudo incontinence (3). Recurrent UTIs can be both a causative factor and a result of LA (2, 3). One patient in our study suffered from recurrent UTIs and was also diagnosed with cystitis cystica. Similar concordance has been reported in two elderly women (6, 7). In children, separation of the labia can be accelerated with topical application of estrogen, and surgery is rarely needed. However, in adult women surgical treatment is usually necessary for symptomatic and severe cases (5). Nonetheless, a recurrence rate ranging from 14 to 20% has been reported following the surgical separation of the labia in prepubertal girls (8).

In our study, we did not encounter any LA relapse during the mean 27-month (range:18-36) follow- up. Based on our experience, several measures can help reduce the recurrence rate of labial fusion. These include placing separate absorbable sutures at the introitus area, minimizing the use of electrocautery, prolonged treatment with topical estrogens, maintaining regular intercourse, and/or using vaginal cones to dilate the vaginal introitus following surgery.

Conclusions

The increasing number of adults LA case reports highlights the importance of conducting a thorough genital examination in all females with hypoestrogenic state and voiding complaints, especially those who are not sexually active. A surgical procedure and proper prolonged application of topical estrogen would serve as suitable treatments for postmenopausal LA. Maintaining regular sexual intercourse or using vaginal cones in selected patients would help decrease the recurrence rate of labial fusion.

Authors' contributions

NMn: Conception and design, critical revision of the manuscript for important intellectual content; FSH: Supervision; ShT: Acquisition of data; MHm: Drafting of the manuscript; AKr: Drafting of the manuscript; NS: Acquisition of data

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Conflict of interest

The author declares that there is no conflict of interest.

Funding

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Ethical statement

The case agreed to report his issue anonymously after signing the informed consent. This case report is based on the CARE checklist.

Data availability

Data will be provided on request.

Abbreviations

LA Labial Adhesion
PVR Post-void Residual Volume
UTI Urinary Tract Infection

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Author (s) biosketches

Mahmoudnejad N, Assistant Professor, Urology and Nephrology Research Center, Shahid Labbafinejad Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
Email: nastaran.mahmoudnejad@gmail.com

Sharifiaghdas F, Professor, Urology and Nephrology Research Center, Shahid Labbafinejad Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
Email: f.sharifiaghdas@gmail.com

Tehrani Sh, Associate Professor, Infectious Disease and Tropical Medicine, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
Email: Tehrani.shabnam89@yahoo.com

Hamidi Madani M, MD, Urology and Nephrology Research Center, Shahid Labbafinejad Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran.
Email: m.hamidimadani70@gmail.com

Karimi Rouzbahani A, MD, Student Research Committee, Lorestan University of Medical Sciences, Khorramabad, Iran.
Email: ariankarimi1998@gmail.com

Saberi N, Assistant professor, Isfahan Kidney Disease Research Center, Isfahan University of Medical Sciences, Isfahan, Iran.
Email: narjessaberi@gmail.com

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