Labial adhesion is a rare condition in reproductive-aged women. There are only a few reported cases of labial adhesion in this period of woman’s life. We herein, present a case of a 22-year-old G1P1 woman with a thick and fibrous labial adhesion. The labial adhesion was excised, and the labial mucosa was sutured under local anaesthesia. Her hormonal profile (FSH, LH and estrogen levels) was found to be normal. The patient was later diagnosed with systemic lupus erythematosus (SLE) by a rheumatologist. It is the first case report that shows a relationship between SLE and labial adhesion.

**KEYWORDS:** Labial adhesion, Labial fusion, Systemic lupus erythematosus.
A more detailed history revealed that oral ulcers had been developing on a recurring basis in the past. She further reported a painless vaginal ulcer which was spontaneously healed in two weeks before experiencing difficulties in sexual intercourse. Considering also the poor obstetric history of the patient, we had brought suspicion upon a connective tissue disorder. The patient was referred to a rheumatologist for a specific evaluation. The immunological panel showed high anti-nuclear antibody (ANA), anti-double-stranded DNA (anti-dsDNA) antibody and anti-Smith (anti-Sm) antibody titers, with low complement three levels. Lupus anticoagulant and anticardiolipin IgG and IgM antibodies were negative. She also declared suffering from skin rashes on her hands developing upon exposure to sunlight. The patient was eventually diagnosed with systemic lupus erythematosus (SLE).

DISCUSSION

Labial adhesion occurs when the labia minora adhere together resulting in a narrowed vaginal opening or introitus. It is most commonly seen in prepubertal girls and postmenopausal women; however, it is an extremely rare condition in women of the reproductive age group. The incidence is estimated to be 0.6-5% in prepubertal girls. The diagnosis is made by the inspection of the vulva. The adhesion typically begins at the posterior fourchette and continues anteriorly. Though it is often an acquired condition, it can also occur as a congenital anomaly, usually accompanied by other genitourinary abnormalities. It can be asymptomatic, especially in prepubertal girls. Anterior adhesions may lead to urinary symptoms such as urinary retention, urinary tract infection, pain or altered urinary stream. The most common complaint in women of the reproductive age group is difficulty in engaging in sexual activity, as was encountered in the present case. Application of topical estrogen creams is the first choice of treatment in prepubertal girls and postmenopausal women but is not usually successful in the reproductive age group. Consequently, surgical division of the adhesion is a better alternative to medical treatment in women of reproductive age. In the present case, we opted for surgical excision of the adhesion because of its thick fibrous nature.

It has been postulated that low levels of estrogen are a predisposing factor for labial adhesion in prepubertal girls and postmenopausal women. In one study, labial biopsies obtained from two patients revealed chronic and acute inflammatory changes, indicating that inflammation may play a role in the formation of labial adhesion in
hypoestrogenic women. However, it may also occur in the presence of normal estrogen levels. In women of reproductive age; female circumcision, herpes simplex, diabetes, pemphigoid, caustic vaginitis, poor hygiene, trauma related to sexual abuse or a fall and lichen sclerosis are reported to be the causes of labial adhesion. In the present case, we excluded hypoestrogenemia as a causative factor since the patient’s estrogen levels were normal. She denied a history of trauma, sexual abuse or vaginal discharge. The patient’s history of preeclampsia, repetitive oral ulcers, painless vaginal ulcer and the presence of labial adhesion arouse the suspicion of systemic lupus erythematosus (SLE) as the underlying cause. With the patient’s consent, detailed investigations were performed. In the following months after surgery, 4 of the 11 American College of Rheumatology criteria required for the diagnosis of SLE; ANA positivity, oral ulcers, photosensitivity and immunological disorder presenting with anti-Sm antibody positivity were documented. It is a rare finding in SLE to have a vaginal ulcerous lesion. We hypothesized that the asymptomatic vaginal ulcer could have caused a chronic inflammatory process eventually resulting in labial adhesion.

To the best of our knowledge, this is the first case report that shows a relationship between SLE and labial adhesion. We believe that SLE should be considered in the differential diagnosis of labial adhesion in women of reproductive age.

Grant Support & Financial Disclosures: None.

Conflict of Interest: The authors have nothing to disclose.

REFERENCES

1. Seehusen DA, Earwood JS. Postpartum labial adhesions. J Am Board Fam Med. 2007;20:408-410. doi: 10.3122/jabfm.2007.04.060214.
2. Kutlu O, Koksal IT. Labial adhesion in a reproductive aged girl. Saudi Med J. 2010;31:202-203.
3. Mayaoglu L, Dulabon L, Martin-Alguacil N, Pfaff D, Schober J. Success of Treatment Modalities for Labial Fusion: A Retrospective Evaluation of Topical and Surgical Treatments. J Pediatr Adolesc Gynecol. 2009;22(4):247-253(247-250). doi:10.1016/j.jpag.2008.09.003.
4. Hettler J. Pediatric and Adolescent Gynecology. In: Fleisher GR, Ludwig S, editors, Textbook of Pediatric Emergency Medicine. Philadelphia. Lippincott Williams & Wilkins. 2010;p 842.
5. Webster JJ, Williams G. Adhesions of the labia minora. Br J Urol. 1996;78(1):146-147.
6. Pulvino JQ, Flynn MK, Buchsbaum GM. Urinary incontinence secondary to severe labial agglutination. Int Urogynecol J Pelvic Floor Dysfunct. 2008;19:253. doi: 10.1007/s00192-007-0415-8.
7. Caglar MK. Serum estradiol levels in infants with and without labial adhesions: the role of estrogen in the etiology and treatment. Pediatr Dermatol. 2007;24:373-375. doi: 10.1111/j.1525-1470.2007.00493.x.
8. Awang NA, Viegas C, Viegas OA. Incomplete bladder emptying due to labial fusion in a pubertal girl: a delayed consequence of female circumcision. Australian and New Zealand J Obstet Gynaecol. 2004;44:372-373. doi: 10.1111/j.1479-828X.2004.00261.x.
9. Hochberg MC. Updating the American College of Rheumatology revised criteria for the classification of systemic lupus erythematosus. Arthritis Rheuma. 1997;40(9):1725. doi: 10.1002/1529-0131(199709)40:9&lt;1725::AID-ART298&gt;3.0.CO;2-Y.
10. Uva L, Miguel D, Pinheiro C, Freitas JP, Marques Gomes M, Filipe P. Cutaneous manifestations of systemic lupus erythematosus. Autoimmun Dis. 2012;2012:834291. doi: 10.1155/2012/834291.

Authors’ Contribution:

DY and MT both involved in manuscript writing.