CASE REPORT

Labial adhesions caused by Stevens–Johnson syndrome

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ABSTRACT

Vulvovaginal sequelae in Stevens–Johnson syndrome have been widely described in published literature, however, there is no consensus regarding effective preventive treatments. Labial minora synechiae as sequelae of Stevens–Johnson syndrome are rare, but can reduce patients' quality of life as a result of dysuria, urinary tract infection, and sexual dysfunction. Occasionally, sequelae can induce adenosis, and subsequently result in malignancy. Therefore, biopsy of the affected tissue in Stevens–Johnson syndrome patients with genital involvement should be considered after the acute phase has been resolved. Here, we report Stevens–Johnson syndrome with labial involvement in a 24-year-old woman who was treated with labial adhesiolysis. We recommend genital examination for all women with Stevens–Johnson syndrome to diagnose genital disease and avoid urinary tract infections and other sexual complications.

Introduction

Stevens–Johnson syndrome (SJS) is a rare and serious condition, belonging to the group of conditions known as toxic epidermal necrolysis. Clinically, patients exhibit cutaneous target lesions, blisters, and epidermal necrolysis associated with hemorrhagic mucositis. In addition to the epidermal layer, lesions may be present in the oral mucosa, esophagus, pharynx, larynx, trachea, vagina, and urethra. If mucocutaneous lesions develop on the face, additional serious ocular symptoms may develop, which may cause blindness. Although genitourinary involvement is not as serious, it may lead to symptoms such as dysuria or an inability to void. Patients with vulvovaginal sequelae of SJS may also complain of dyspareunia and postcoital bleeding. Subsequent pathologic changes in vulvovaginal lesions, such as adenosis, are also commonly reported in patients with SJS. We report the case of a woman with dysuria, vulvar irritation, and labial adhesion after an episode of SJS, and provide a literature review.

Case report

A 24-year-old female patient presented to the Obstetrics and Gynecology Department (Busan Paik Hospital, Busan, Korea) with dysuria, vulvar irritation, and labial adhesion. The patient had a history of SJS secondary to the administration of medication prescribed to relieve cold symptoms in Vietnam, but had no other remarkable medical or surgical history. The patient reported that the lesions appeared within a few hours of taking the medication, which contained acetaminophen. According to the patient, she observed a marked, irregular purpuric macular rash, involving mainly her trunk and limbs, along with conjunctivitis and oral ulceration, leading to the development of multiple fluid-filled purple lesions. On physical examination, erythematous maculopapular lesions were observed over her entire body, along with congestion and ocular swelling. Diffuse oropharyngolaryngeal swelling and injection were also observed during laryngoscopy. During the acute phase, the patient was treated with intravenous dexamethasone, which was subsequently changed to oral prednisolone in the Department of Dermatology of our hospital. Topical application of antibiotics and dexamethasone for keratoconjunctivitis was prescribed, along with corticosteroid lotion for facial and cutaneous lesions. The patient responded well to treatment, and had noticeably improved after 10 days. She remained afebrile, and a generalized subsidence in her rashes was observed, along with reduced oral swelling. The lesions took approximately 1 month to heal completely.

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One month later, the patient again visited our department complaining of urinary difficulty and vulvar irritation. On vulvovaginal examination, labial adhesion was observed. The external urethral orifice was not visible, since the bilateral labium minora had adhered together. The vaginal introitus was very narrow due to the labial adhesion observed in the first visual inspection of the vulva. However, the labium minora partially separated during the vaginal examination. We observed that labial fusion was likely the cause of the patient’s urinary difficulty and dysuria (Figure 1A and B). Labial adhesion was separated by incision with electrocutting, and the labial mucosa was sutured to reconstruct the labium minora and free the clitoris. Then, each labial minora was fixed on bilateral labial majora with stay suture using 3-0 nylon (Ethicon Inc, Somerville, NJ, USA) to prevent readhesion. Total suture length was approximately 1.5 cm and povidone–iodine dressing was performed after wound closure. For the differential diagnosis of vulvovaginal adhesion, the separated labial tissue was sent for pathological analysis that showed chronic inflammation of the labial mucosa. The patient was discharged the day after surgery.

Discussion

Although SJS frequently affects the genitalia, little consensus exists regarding effective prevention strategies. Labial sequelae such as labial fusion and vaginal opening stenosis reportedly occur in 18–28% of patients.1,6 If mild labial adhesion is involved, patients may be asymptomatic, although they may experience symptoms such as urinary tract infection, dysuria, and dyspareunia. The correlation between the severity of SJS and genital complications is not well known, although these are considered the most severe systemic symptoms, with the highest potential for sequelae.

SJS commonly occurs in children and young adults, with Ting and Adam7 reporting that 14 patients (14%) had genital ulceration during the acute phase of balanitis or vulvovaginitis, which subsequently resolved. De Jesus et al.2 reported the case of a 10-year-old girl who presented with abdominal pain, dysuria, and extensive labial synechiae 6 years after SJS was treated with surgical incision for labial adhesion along with suture of the labial mucosa to reconstruct the labia and clitoral prepuce. If genital sequelae after SJS occurs before menarche, and vaginal adhesion is not severe, the symptoms of the adhesion may be few.

However, postmenarcheal patients experience several symptoms, including amenorrhea, cyclic abdominal pain, dysuria, and dyspareunia due to labial adhesion or introitus stenosis. Vaginal adenosin and endometriosis after SJS have also been reported.2,6 De Jesus et al.2 have offered a possible explanation for this phenomena of vulgar or vaginal adenosin and endometriosis or synechiae after SJS, whereby the vaginal or vulvar necrosis in opposing walls heal by adhesion. As a result, the tubal or uterine epithelium can implant in the new location during SJS. Therefore, biopsy of genital synechiae should be performed to confirm adenosin. In our case, the fused labia were separated by incision, and tissue from the labial mucosa was sent for differential diagnosis of adenosin by pathology.

Women with a history of SJS should be examined to determine vulvovaginal involvement and the presence of genital sequelae. Therefore, when female patients with SJS are treated for systematic lesions, gynecological consultation is necessary for genital examination. Prompt examination may facilitate early detection and management of genital complications as well as prevent urinary tract infection or endometriosis, thus improving quality of life.

References