

Lichen sclerosus causing obstructive urinary retention and pyocolpos: A case report

Michelle Van, Georgina Davis

ABSTRACT

Introduction: Lichen sclerosus is one of the most common dermatological conditions affecting the vulva, perineum, and perianal area. The etiology is not entirely understood but appears to be multifactorial and most commonly affects women above the age of 50. Lichen sclerosus can cause extreme pruritis, pain, and loss of vulval architecture with resorption of the labia minora and stenosis of the vaginal introitus.

Case Report: An 82-year-old woman presented to the Emergency Department with lower abdominal pain and dysuria. She was found to be in urinary retention. On insertion of an indwelling catheter, severe anatomical distortion and scarring of the vulva was noted with fusion of the labia majora in the midline, stenosis of the vaginal orifice, and excoriated skin. A computed tomography (CT) scan of the abdomen and pelvis was performed as her pain persisted despite bladder emptying and was significant for a pyocolpos with a dilated vagina and ring enhancement. Examination under anesthesia and biopsies confirmed severe lichen sclerosus resulting in obstructive pyocolpos and urinary retention. The patient was commenced on ultrapotent topical steroids and remains under clinical surveillance.

Conclusion: This case illustrates a rare and important complication of untreated lichen sclerosus.

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INTRODUCTION

Lichen sclerosus is a common and chronic skin condition that affects the vulva, perineum, and perianal areas. It most commonly affects postmenopausal women; however, there is a peak incidence in pre-pubertal girls [1]. The gold standard for diagnosis is by histopathological examination of a biopsy and treatment is by topical potent or ultra-potent steroids [2]. Lichen sclerosus typically causes itching and pain and gradually leads to scarring and adhesions of the non-hair bearing skin of the vulva [3]. The condition carries a 5–7% risk of squamous cell carcinoma (SCC) of the vulva [4], therefore, accurate diagnosis, appropriate treatment, and follow-up are important.

There is a paucity of literature on the structural and obstructive urinary retention complications of lichen sclerosus in women, while the condition is well described in men [5, 6]. Furthermore, pyocolpos, a complication of hydrocolpos, is an entity rarely seen beyond infancy and adolescence and is due to the retention of secretions in the vagina in the presence of an imperforate hymen [7], transverse vaginal septum or obstructed hemivagina in cases of Müllerian abnormality [8–10].

We describe a case of urinary retention and pyocolpos as a result of obstruction from severe lichen sclerosus.

CASE REPORT

An 82-year-old woman presented to the Emergency Department with gradually increasing lower abdominal pain over the past three weeks, associated with dysuria for which she had received several courses of oral antibiotics from her General Practitioner (GP). In the days preceding her presentation with acute pain she had been finding it increasingly difficult to pass urine and described poor flow and sensation of incomplete bladder emptying. The patient did not describe issues post-menopausal bleeding or abnormal vaginal discharge however had periods of severe vulval itch. Her past medical history was significant for ischemic heart disease with two recent cardiac stents, hypertension, hemochromatosis, osteoporosis, and fibromyalgia.

On examination, the patient was afebrile and her vital signs stable. Her abdomen was soft with suprapubic tenderness and a point of care bladder scan suggested a retained volume of 400 mL. On insertion of an indwelling catheter, she was noted to have abnormal vulval anatomy. The labia majora were fused in the midline with a small caliber introitus. There were areas of excoriation and white hypertrophic skin surrounding the vulva, perineum, and perianal areas. The clinical impression was severe lichen sclerosis.

A CT abdomen pelvis was performed as the patient had ongoing abdominal pain despite the bladder being emptied with an indwelling catheter. The CT was significant for a dilated vagina with ring enhancement suggestive of pyocolpos (Figure 1). There was no dilatation of the endometrial canal and a small amount of free fluid in the pelvis. Blood tests demonstrated mildly elevated white cell count $11.1 \times 10^9/L$ and normal C-reactive protein (CRP) 1.4 mg/L. The remainder of her blood tests was normal. Her urine dipstick was suggestive of a urinary tract infection and sent for culture and sensitivities.

Examination under anesthetic was performed. At the time of positioning in theatre, she was found to have a turbid bloody fluid spontaneously draining from the introitus. The labia majora were approximately 90% fused and the urethra was unable to be visualized externally. There was a 7 mm vaginal orifice. There were no obvious areas concerning for invasive malignancy (Figure 2). A vaginoscopy identified hyperemic vaginal wall tissue and a very atrophic cervix. There was no evidence of vesico-vaginal fistula. The vaginal introitus was gently dilated to a Hagar size 14 and a cytology brush passed for liquid based vaginal cytology. Targeted vulval biopsies were taken and submitted for histopathological examination.

The vaginal liquid-based cytology was showed no evidence of squamous epithelial lesion or malignancy. The low vaginal swab and culture of the pyocolpos was sterile. The vulval biopsies confirmed established lichen sclerosis with lichenification, squamous hyperplasia with no evidence of vulval intraepithelial neoplasia (VIN) or malignancy. A diastase resistant periodic acid schiff (DPAS)

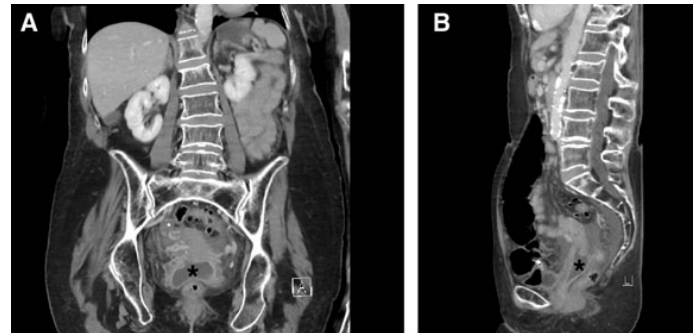


Figure 1: Coronal and sagittal CT images documenting pyocolpos. (A) Coronal CT image illustrating ring enhancing dilated vagina in keeping with pyocolpos (*). (B) Sagittal CT image documenting pyocolpos (*).



Figure 2: Intraoperative photograph of lichen sclerosis, showing extensive destruction of the vulval anatomy, fusion of the labia majora, stenosed vaginal orifice and widespread excoriation and lichenification.

stain on the biopsies was negative for superimposed fungal elements.

Following the procedure, the patient's abdominal discomfort significantly improved and after the removal of her catheter had a successful trial of void. She was continued on oral antibiotics for five days and did not develop any urinary symptoms. The patient was commenced on topical

Betamethasone Dipropionate 0.05% (Diprosone OV Ointment) and remains under clinical review.

DISCUSSION

Lichen sclerosus is a chronic dermatological condition with peak incidences in pre-pubertal girls and more commonly, post-menopausal women [1]. The etiology is unknown but is thought to be multifactorial with genetic factors, hormonal influences, and an association with autoimmune disorders [11]. Its symptomology is characterized by itching and irritation of the vulva and on examination, demonstrates white and thin papules which may coalesce to form plaques and sometimes also affect the perineum and anus in a “figure of eight” formation [3]. Excoriation can cause lichenification of the skin and relatively mild contact through rubbing or intercourse can cause petechiae [3]. The cervix and vagina are usually spared however rarely, in significant vaginal prolapse with keratinization, the vaginal mucosa can develop disease [12, 13]. Extra-genital lichen sclerosus can also occur in 10% of cases, commonly affecting the upper trunk, axillae, buttocks, and lateral thighs [1].

In untreated disease, the scarring process of lichen sclerosus can cause the distinction between labia minora and majora to be lost, labial agglutination, for the fused prepuce to obscure the clitoris and for the introitus and perineum to shrink or stenose, causing dyspareunia [14]. These complications can also be observed in other vulval dermatoses including lichen simplex and vulval psoriasis [15, 16]. With advanced disease, the labia can be fused such that there is only a small posterior vaginal orifice [1]. Rarely, with extensive fusion, obstruction of the urethra and vaginal canal can occur [14].

Patients with lichen sclerosus are also prone to infection from organisms such as *Candida*, *Staphylococcus aureus*, and herpes simplex [14]. Patients on estrogen and progesterone replacement, or those with concurrent diabetes are particularly at risk of superimposed candida infection [17]. There was no evidence of fungal elements in the pathology specimens from our case.

Untreated lichen sclerosus can also cause malignant changes to the vulval epithelium with a cumulative risk of approximately 6.7% [11]. Squamous cell carcinoma of the vulva in the older patients is associated with lichen sclerosus [as opposed to human papillomavirus (HPV) in younger patients with SCC] and 60% of vulval SCC have been found to have background lichen sclerosus on histopathology [4, 18]. Despite not affecting the vaginal mucosa in most cases, lichen sclerosus has also been shown to be associated with increased risk for vaginal cancer [19].

Fortunately, with appropriate treatment and disease control, the risk of malignancy and loss of vulval architecture can be reduced [20]. Topical steroids and emollients are the mainstay of treatment however problems with vulvodynia and psychosexual issues require multidisciplinary care, and in cases of severe deformity

causing urinary symptoms, surgical intervention may be required [21]. This can involve the surgical division of adhesions, perineotomy, or skin flaps to restore normal anatomy however risks recurrence of disease and deformity [22, 23]. Patients with lichen sclerosus, even with good disease control, will require lifelong monitoring of their symptoms and regular self-examination due to the possibility of recurrence and the risk of SCC [4].

A focused vulval history was sought after the clinical impression of lichen sclerosus was made in this patient. Throughout her life she had suffered severe superficial dyspareunia, vulvodynia, and described periods of severe itch. Her last successful cervical screening test was in her 30s. A short course of topical estrogen was prescribed to the patient in her 50s to achieve a further cervical screening test; however, this was unsuccessful and the treatment discontinued due to patient perceived risks of breast cancer. It is likely that these long-term symptoms represented emergence and progression of the disease over many decades subsequently resulting in a disengagement from routine gynecological screening due to symptoms of vulval discomfort. This represents a missed opportunity for vulval inspection and timely diagnosis and management—a chicken and egg scenario.

In this case, progression of disease and significant labial fusion caused a mechanical obstruction to the passage of vaginal secretions and the external obstruction of the urethral may have also caused retrograde flow of urine into the vagina—leading to the patient’s abdominal pain and presentation with pyocolpos. The significant size of the pyocolpos had also caused acute urinary retention due to urethral compression which was relieved following drainage. Given the severity of the changes in vulval architecture, it is unlikely that the patient will be able to achieve normal appearance of the vulva with topical therapy alone, however treatment goals will be to prevent further episodes of urinary retention and pyo or hydrocolpos. Intensive gynecological follow-up is in place to monitor the response to treatment and mitigate any malignant transformation.

CONCLUSION

This case is an interesting presentation of acute abdominal pain in an 82-year-old woman. The underlying mechanism for her presentation was urinary retention from apparent obstructive pyocolpos from severe lichen sclerosus. This case illustrates the importance of addressing women’s gynecological health throughout their lifetime.

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Author Contributions

Michelle Van – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Georgina Davis – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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