

# Plasma cell vaginitis and cervicitis

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## ABSTRACT

**Introduction:** Plasma cell vulvitis (PCV), also known as Zoon's vulvitis, is a rare, chronic inflammatory disease characterized by erythematous plaques with a red, orange hue and increased plasma cells typically affecting the vulva. **Case Series:** This case series details two clinical presentations of plasma cell mucositis affecting the vagina and cervix, resulting in copious, purulent vaginal discharge. Vaginal and cervical histopathologic findings of a plasmacytic infiltrate confirmed the diagnosis of plasma cell mucositis. Both women were successfully treated with a vaginal estradiol/hydrocortisone/clindamycin cream and experienced complete resolution of symptoms and physical exam findings. **Conclusion:** Plasma cell mucositis may be present on the vulva, vagina, and cervix.

**Keywords:** Cervicitis, Plasma cell mucositis, Plasma cell vulvitis, Vulvovaginitis, Zoon's vulvitis

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## INTRODUCTION

Plasma cell vulvitis (PCV), also known as Zoon's vulvitis, is a rare, chronic inflammatory disease characterized by erythematous plaques with a red, orange hue and increased plasma cells affecting the vulva [1, 2]. This condition has also been found to affect other orificial tissue, including the mouth and upper digestive tract [3, 4], called plasma cell mucositis. There have been only three documented cases of plasma cell vaginitis and two reported cases of plasma cell cervicitis [5–8]. This case series details two clinical presentations of histology-confirmed plasma cell mucositis affecting both the vagina and cervix, providing further evidence of plasma cell vaginitis and cervicitis.

## CASE SERIES

### Case 1

A 64-year-old, post-menopausal woman presented with a six-year history of copious, yellow vaginal discharge. For the past five years, she had been managing her menopausal symptoms with topical testosterone 1% 0.5 mg QD and an estrogen patch 0.0375 mg/QD twice weekly. She had tried the following treatments for the vaginal discharge without significant relief: oral metronidazole, azithromycin, topical metronidazole, fluconazole, topical estrogen, and clindamycin. Physical

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examination of the vulvar vestibule and urethral meatus revealed erythematous papules that were severely tender (Figure 1). Copious, yellow discharge was present in the vaginal vault with evidence of erythematous papules on the vaginal mucosa. The vaginal pH was 5.7 and many white blood cells (WBCs) and parabasal cells were noted on saline microscopy. Many small erythematous papules were noted on the cervix (Figure 2). A cervical biopsy was obtained with Tischler biopsy forceps. Histology showed cervical squamous epithelium with a lichenoid lymphoplasmacytic infiltrate (Figure 3), confirming a diagnosis of plasma cell cervicitis. She was instructed to vaginally insert 1 gram of estradiol 0.02%/hydrocortisone 10%/clindamycin 2% daily for four weeks, every other day for four weeks, then twice per week. After three months, she experienced complete resolution of her leukorrhea, microscopy of her vaginal discharge was normal with a pH of 4.5, and she had resolution of the erythema and papules on the vulvar vestibule, urethral meatus, and cervix.

## Case 2

A 35-year-old, pre-menopausal woman presented with copious, yellow vaginal discharge for the past 15 years. Due to the volume of her discharge, she changed panty-liners several times per day. She tried the following treatments without significant relief: topical and oral



Figure 2: Erythematous papules on the vaginal wall.

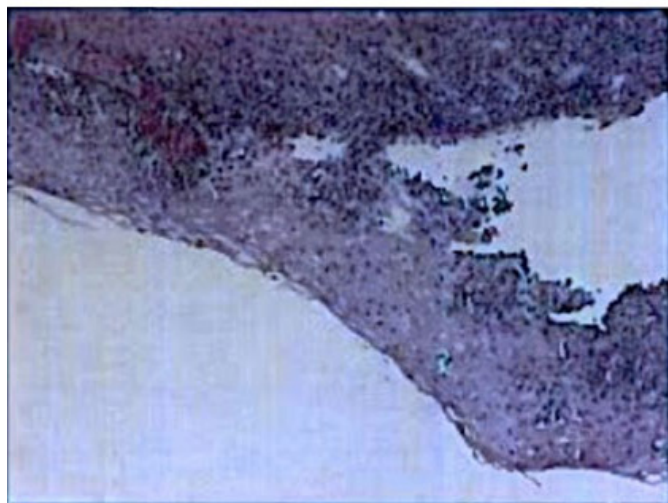


Figure 3: Cervical squamous epithelium with lichenoid inflammatory infiltrate consisting mainly of lymphocytes and rare plasma cells.



Figure 1: Erythematous papules on the vulvar vestibule and urethral meatus.

metronidazole, miconazole, azithromycin, levofloxacin, valacyclovir, ciprofloxacin, doxycycline, fluconazole, probiotics, and oral and vaginal clindamycin. Upon examination, multiple erythematous papules were visualized on the urethral meatus, vagina, and cervix. There were no papules noted on the vulvar vestibule. Copious, yellow discharge was appreciated in the vaginal vault. Saline microscopy showed a pH of 5.5 with many WBCs and parabasal cells. A vaginal biopsy was obtained to confirm the clinical diagnosis of plasma cell vaginitis. Histology showed mucosal hyperplasia with underlying dense lymphoplasmacytic lichenoid infiltrate, confirming a diagnosis of plasma cell vaginitis. The patient was instructed to vaginally insert 1 gram of estradiol 0.02%/

hydrocortisone 10%/clindamycin 2% daily for six weeks, then decrease to every other day. At follow-up, she had complete resolution of her vaginal discharge, microscopy was normal (no WBCs or parabasal cells) with a pH of 4.0, and the urethral, vaginal, and cervical papules had completely resolved.

## DISCUSSION

In 1952, Professor Johannes Jacobus Zoon described Zoon's balanitis, an inflammatory condition which causes increased plasmacytic infiltrate of the penis and prepuce [1]. Zoon's vulvitis, commonly referred to as PCV or vulvitis circumscripta plasmacellularis, was first documented by Garnier two years later [1, 2]. Plasma cell vulvitis is a rare, chronic inflammatory disease characterized by erythematous plaques with a red, orange hue affecting the vulva. Plasma cell vulvitis predominantly affects the vulvar vestibule, but may extend to the labia minora and majora. Plasma cell mucositis is an inflammatory ulcerative condition of the mucous membranes of the oral cavity and upper aerodigestive tract defined by erythematous ulceration and a plasmacytic infiltration of the mucosa [3, 4].

The etiology of PCV and mucositis is unknown. There is speculation that PCV may be caused by an autoimmune disorder due to associations with desquamative gingivitis, autoimmune polyglandular endocrine failure, and circulating immune antibodies [8]. Common symptoms of PCV include vulvar pruritus, burning, pain, and dyspareunia [2]. Proper diagnosis of PCV and mucositis requires clinical examination and histology confirming the presence of dense plasma cellular infiltrate [9].

Due to the rarity of the disease, lack of research, and inconsistent response to treatment, there is no standard treatment regimen for PCV. However, many different treatments have been utilized for PCV, including topical corticosteroids, topical clindamycin, topical misoprostol, topical tacrolimus, intralesional interferon- $\alpha$  injections, topical imiquimod, topical estrogen, topical cyclosporine, antifungals, cryotherapy, laser ablation, antibiotics, fulguration, and surgical resection [1, 2, 9]. Currently, most widely accepted treatment is topical corticosteroids or calcineurin inhibitors [10].

There have been only a few documented cases of plasma cell vaginitis and plasma cell cervicitis [5–8]. In two of the previously documented cases of plasma cell vaginitis, the patients improved symptomatically when treated with topical fusidic acid cream or intravaginal cinchocaine hydrochloride/hydrocortisone suppositories [5, 6]. In the third case, the vaginal tissue demonstrating plasma cell mucositis was excised during surgical treatment for pelvic organ prolapse and urinary stress incontinence [7]. In the case of plasma cell cervicitis, the diagnosis was confirmed on pathology of the hysterectomy specimen [8].

In the current case study, both patients had erythematous plaques on the urethral meatus, vagina,

and cervix. Only one of the patients had vulvar involvement. Vestibular and urethral meatus biopsies were not performed and therefore the urethral meatus and vestibular erythema and tenderness could have been due to PCV or irritation caused by the inflammatory leukorrhea. Desquamative inflammatory vaginitis (DIV) is persistent vaginitis with similar symptoms as these two documented cases. It is characterized by a purulent vaginal discharge causing vulvovaginal irritation. Diagnosis is based upon an elevated vaginal pH with increased number of leukocytes and parabasal epithelial cells noted on saline microscopy. The etiology of DIV is still unknown [11]. Plasma cell vaginitis and/or cervicitis may potentially be a cause of DIV. Vaginal and cervical biopsies confirmed the diagnosis of plasma cell vaginitis and cervicitis. Both patients improved symptomatically and clinically after treatment with intravaginal estradiol 0.02%/hydrocortisone 10%/clindamycin 2%. Considering success of treatment in these cases, as well as in the aforementioned case studies of plasma cell vaginitis, an intravaginal corticosteroid appears to be a key component of treatment.

## CONCLUSION

Plasma cell mucositis may be present on the vulva, vagina, and cervix. Plasma cell vaginitis and cervicitis may result in a copious, yellow vaginal discharge. The diagnosis is confirmed by biopsy with a plasmacytic infiltrate on histology. Intravaginal estradiol 0.02%/hydrocortisone 10%/clindamycin 2% may be an effective treatment for plasma cell cervicitis and vaginitis.

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**Author Contributions**

Leia S Mitchell – Conception of the work, Design of the work, Acquisition 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

Karissa Barela – Design of the work, Acquisition of data, 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

Jill M Krapf – Design of the work, Acquisition 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

Vaishnavi Govind – Design of 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

Hillary Tolson – Acquisition of data, 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

Andrew T Goldstein – Conception of the work, Design of the work, 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

**Guarantor of Submission**

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**Conflict of Interest**

Leia Mitchell, Hillary Tolson, Vaishnavi Govind, and Jill Krapf report no conflicts of interest. Andrew Goldstein is a consultant to SST, Dare, Ipsen, Grunenthal and has received research funding from Elen, Ipsen, Endoceutics, Grunenthal, Dare, and SST.

**Data Availability**

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

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