



Case Presentation
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A Rare Case of Vulvitis

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Abstract

Introduction: Zoon vulvitis is a rare benign chronic inflammation of the vulva which affects women of various ages. Because of its rarity and atypical appearance resembling many other vulvar conditions, diagnosis is often difficult and delayed. Differential diagnosis is made with a wide range of vulvar lesions. Final diagnosis is histological, vulvar biopsy revealing a plasmacytic infiltrate. Literature on this subject is scarce and only few systematic reviews are published. The condition is often refractory to treatment and every case presentation adds new interesting information on this topic.

Case report: We report a case seen in our department. A 62-year-oldpostmenopausal woman presented with vulvar pruritus and pain, lasting for more than6 months. Local examination revealed well-defined brick-like red erythema and atrophy involving the introitus and labia minora. Fungal

and viral infections were excluded by performing specific laboratory tests. Biopsy was performed and revealed specific dermal infiltration with plasma cells, haemosiderin deposits and vessel dilation, which are characteristic to Zoon vulvitis. We tried several treatment options, with variable outcomes, trying to find the best solution for our patient.

Conclusion: Although plasma cell vulvitis is a benign inflammatory condition of the vulvar mucosa, its clinical manifestations are misleading because many vulvar dermatoses overlap. Diagnosis and symptom relief are often delayed with months or years because of patient embarrassment in seeking treatment and because clinicians are unaware of the condition.

Keywords: Zoon vulvitis; Plasma cell vulvitis

Introduction

First described by Zoon in 1952, Plasma Cell Vulvitis (PCV) or vulvitis circumscripta

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plasmacellularis is a rare idiopathic dermatosis of the vulva. The histologic aspect was previously described in men (plasma cell balanitis) and it is thought to be part of a spectrum of plasma cells mucositis which can also affect oral mucosa and glans mucosa [1]. In women, PCV is a benign, chronic condition found mostly in postmenopausal women, but few cases were reported in pediatric patients as well. Its etiology is unknown, literature studies suggesting autoimmune causes, but also hormonal, infectious, and irritant factors [2]. The clinical appearance shows a well circumscribed brick-red, glistering erythema and atrophy of the labia minora, the introitus, vestibule, clitoris and labia majora. Patients may have erosions and ulcerations, extended or solitary lesions [3]. They can be asymptomatic, but patients often present with itching, burning and discomfort. Some patients report dysuria, dyspareunia, vulvar pain and leucorrhea. Differential diagnosis is made with lichen planus, chronic candidiasis, mucous membrane pemphigoid, contact dermatitis and rarely, vulvar cancer. Vulvar biopsy is the only procedure that ensures a final diagnosis [4].

Histopathologic findings include inflammatory infiltrate with plasma cells, diamond-shaped keratinocytes, haemosiderin deposition and vessel dilation with erythrocyte extravasation. Coexisting conditions were rarely found in patients with PCV, but studies report an increased incidence of autoimmune diseases: cutaneous lupus erythematosus, autoimmune thyroiditis, adrenal insufficiency, rheumatoid arthritis. Other associated vulvar conditions found in patients with PCV were vulvar lichen sclerosus, candidiasis, human papilloma virus infection, herpes simplex virus infection, vulvar allergic contact dermatitis, asthma, allergies [5]. A systematic review on PCV, by Sattler et al. (2021) did not find any cases of PCV progressing to differentiated vulvar

intraepithelial neoplasia. According to the same review, which comprised a total number of 196 patients, only three cases of PCV were reported to develop from mucinous metaplasia [6]. The most common treatment option described in literature was topical corticosteroids and immunosuppressants. Some studies also report adding topical combinations of fusidic acid, betamethasone and tacrolimus. The use of topical hydrocortisone, imiquimod, estrogen, clindamycin, other antibiotics and antifungal ointments was also reported, with variable outcomes. Some clinicians also used systemic corticosteroids, oral antibiotics, oral estrogen, oral progesterone, acyclovir. Other treatment modalities described in literature included excision, fulguration, CO2 laser therapy and Platelet-Rich Plasma (PRP) [1,6].

Case Presentation

We report the case of a 62-year-old postmenopausal woman, who presented to the gynecologist for vulvar itching, dysuria and the occurrence of red lesions on the internal side of one of her labia, which extended symmetrically to the other labia as well in the past months. The symptoms appeared more than six months prior to her seeking medical help. The patient's medical history did not show any associated pathologies. The patient could not make any association between vulvar symptoms and a possible allergic reaction or use of local ointments.

The local examination revealed vulvar atrophy and brick-red like well-defined lesions on both labia, extending towards the clitoris and introitus, with a symmetrical appearance. The vagina and cervix appeared normal, with no pathological leucorrhea (Figure 1).



Figure 1: Symmetrical, shiny, brick-red lesions of the vulva and atrophy.

A Pap smear and swabs were taken to exclude chronic fungal and viral infections. The Pap smear appeared negative for intraepithelial lesions or malignancy, showing only vulvar atrophy. No fungal, viral or bacterial infections were found. The patient was also referred to a plastic surgeon for an extended medical opinion and a vulvar biopsy was taken to have a clear diagnosis and to exclude

lichenplanus, lichen sclerosus, other types of dermatoses and neoplasia.

The histopathological examination revealed erosion of the vulvar mucosa overlying a dense stromal plasma cell infiltrate. The infiltrate contains numerous plasma cells and rare eosinophils. Vascular ectasia and extravasated erythrocytes are also seen (Figure 2-4).

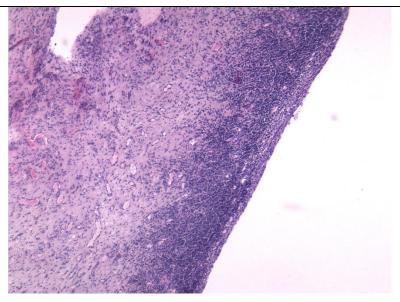


Figure 2: Zoon vulvitis. Note the attenuation and erosion of the vulvar mucosa overlying a dense stromal plasma cell infiltrate. HE stain 10x.

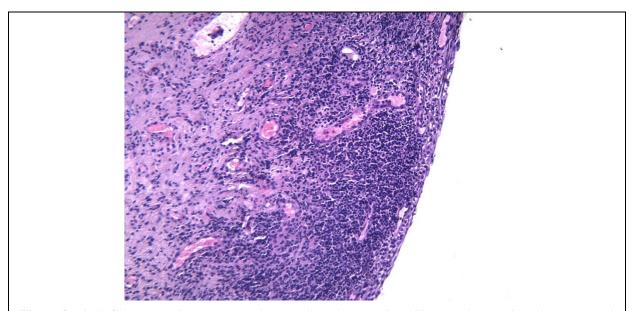


Figure 3: The infiltrate contains numerous plasma cells and rare eosinophils. Vascular ectasia and extravasated erythrocytes are also seen. HE 20x.

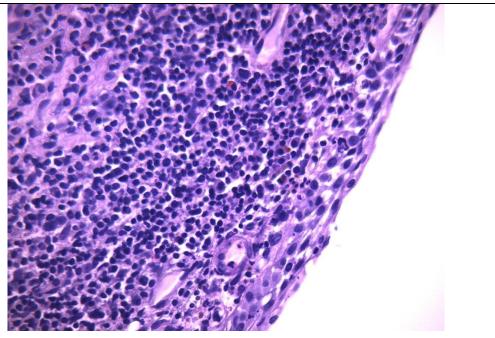


Figure 4: High power magnification highlighting lack of stratum granulosum and stratum corneum layers and mild spongiosis. HE stain 40x

The final diagnosis was plasma cellular vulvitis (Zoon vulvitis) and atrophy. The patient received a topical estrogen cream (promestriene) for a month. Because the results were unsatisfactory, we added topical Tacrolimus 0.03% applied daily for another 30 days.

After two months of local treatment, the patient returned for a follow-up. Local inspection showed a slightly improved aspect of the lesions, with a less intense orange coloring, less contouring, but with persistent erosions on the right minor labia (**Figure 5**).



Figure 5: A slightly improved aspect of the vulvar lesions after 2 months of topical estrogen and tacrolimus 0.03%.

Vulvoscopy revealed leukoplakia on the right labia minor, therefore a new biopsy was indicated in the future to exclude precancerous or cancerous lesions (**Figure 6**).



Figure 6: Lekoplakia on the left labia seen at vulvoscopy after applying acetic acid.

We decided to change the treatment, starting Tacrolimus 0.1% ointment and topical mometasone furoate, but the patient did not tolerate the

corticosteroid, reporting severe burning and therefore mometasone furoate was discontinued. The patient continued with a combination of local estrogen and tacrolimus, applied twice a day for at least 3 months.

Discussion

Zoon vulvitis is a rare idiopathic, chronic and benign inflammation of the vulva. The disease affects women between 8 and 70 years old, as described by literature studies, with a higher prevalence in postmenopausal patients. Etiology is possibly linked to a reaction of the vulvar mucosa to infection or trauma, but an autoimmune response may also be involved. Vulvar lesions mimic other vulvar conditions that need specific treatment, so a correct diagnosis is essential. Most patients are asymptomatic or describe vulvar itching and pain, associated with well-defined, orange-red, shiny plaques on labia minora, with a symmetrical and bilateral distribution and a tendency to extend and converge. The lesions persist for months or years and often reappear. Diagnosis is made by examination which histological reveals inflammatory infiltrate with >50% plasma cells, vascular proliferation, haemosiderin depositions and extravasation of erythrocytes. Histology helps differentiate Zoon vulvitis from other vulvar conditions including squamous cells carcinoma [1,6-8]. Treatment options of Zoon vulvitis are scarce, with variable clinical outcomes. Most clinicians use topical high potency corticosteroids and tacrolimus, but other options are antifungals, estrogen, surgical excision, laser [9]. Vulvar health may be improved by newer treatment options like PRP. Unlike Zoon balanitis, there is no report of malignant changes is Zoon vulvitis, although some cases with moderate dysplasia have been noted [10]. We described the case of a postmenopausal woman who presented with vulvar itching and extended vulvar lesions, who persisted for more than 6 months before she was referred to the department of Obstetrics and Gynecology. After we

consulted with a plastic surgeon and dermatologist, we performed a vulvar biopsy which revealed the final diagnosis of plasma cell vulvitis. We tried several local treatment options, starting with topical estrogen, considering that estrogen deficiency is the main cause of vulvar atrophy in menopause. Our patient had a slight improvement of her condition after one month and was unsatisfied with the outcome. Therefore, after reading more studies, which described better results with increased concentrations of tacrolimus and topical corticosteroids, we decided to increase the concentration of tacrolimus from 0.03% to 0.1% and to add a topical corticosteroid. The corticosteroid ointment was discontinued because the patient did not tolerate it. We recommended the patient to continue with local Tacrolimus 0.1% and estrogen ointment for at least 3 months. Furthermore, because histological examination is the only one who can diagnose any suspicious lesion, we consider taking a new biopsy in the future.

Conclusions

Plasma cell vulvitis is a rare type of vulvar disease, often under recognized because many patients delay seeking for medical help because of shame or stigma and because most clinicians are unaware of this pathology. It should be considered in patients with vulvar erythema, burning sensation and a predominant plasma cell infiltrate in histology. Many patients are seen by several specialists before receiving an adequate diagnosis and treatment. In our case, after trying topical estrogen and local corticosteroid alone, we noted an improvement after using a combination of topical tacrolimus 0.1% and local estrogen, therefore, we advised the patient to apply this combination for an extended period, hoping for better outcomes. We also advised our postmenopausal patient to have regular follow-ups and consider a new biopsy. It is important to note that Zoon vulvitis is a chronic disease, with no standard treatment, with a variable response and a longer time needed to obtain a favorable outcome. We presented this case report to raise awareness about this rare, under recognized pathology.

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