Vulvodynia: a case series of a poorly recognized entity

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Summary

Vulvodynia remains a poorly recognized entity with unclear pathogenesis. In a case series of six patients with vulvodynia over a five-year period in a tertiary university hospital, we describe the clinical features, the diagnostic procedures, the impact on each patient's emotional status and discuss the necessity and efficacy of the chosen treatment options in accordance with the current therapeutic guidelines.

Key words: Vulvodynia; Chronic vulvar pain.

Introduction

Vulvodynia is an uncommon and poorly recognized entity with unclear pathogenesis. In a small case series of six patients with vulvodynia over a five-year period at a tertiary university hospital we describe the clinical features, the diagnostic procedures, family and professional parameters, the impact on each patient's emotional status and discuss the necessity and efficacy of the chosen treatment options in accordance with the current therapeutic guidelines.

Case Series

Case 1

A 48-year-old patient, married, para 0, gravida 0, presented with dysesthesia, dyspareunia and burning sensation of the vulva. The symptoms had exacerbation and remission of various duration depending on stress-associated factors. She was examined by a number of different physicians and was thoroughly investigated with no pathological results. Topical antifungals, topical steroids and tranquillizers were administered resulting in no relief of the symptoms. By searching her history, it was revealed that the patient had a neurotic and hypochondriac personality long before the vulvodynia signs presented. On clinical examination, there was slight erythema of the vulva, while the vagina and cervix were normal. When pressing the vestibulus with a cotton swab (Q-tip test), hypersensitivity and a burning sensation were induced. Having found no etiological relations, the diagnosis of the vulvodynia was set and an explanation of the unclarified nature of this entity was given to the patient. One year later, symptoms were totally absent and after a five-year follow-up, the patient remains symptom-free. Meanwhile a severe family issue, probably the major cause of the patient's stressed condition, had been resolved.

Case 2

A 52-year-old patient, married, gravida 1, para 1, presented with tense dyspareunia, dysuria and typical burning sensation of the vulva. Symptoms were so severe for more than three years, that her sexual life and her emotional world were heavily disturbed. The patient had visited many different physicians, undergone a number of laboratory examinations and was treated with various regimens. On clinical grounds, there was no lesion on the vulva, vagina or cervix. Having a long talk with the patient, no preceding chronic stress causative facts in the family or at her work environment were revealed. The Q-tip test was positive. Biopsies from three different sites of the afflicted area were taken showing findings of a non specific chronic inflammation. After a thorough explanation of the nature of vulvodynia, in the absence of other treatment modalities, laser ablation was applied in order to reduce the number of sensory nerve fibres. Ten months later the patient presented in the same condition.

Case 3

A 38-year-old patient, married, gravida 1, para 1, presented with a three-month duration of intense burning and pain sensation on the clitoris area. The Q-tip test was positive, although the symptoms were present with or without any pressure. On clinical examination the area was normal, with no erythema or edema. A number of physicians had already examined her resulting in no diagnosis. Laser ablation below the clitoris area was then performed with no response. This discomforting entity began to influence her personality, resulting in depression. She was treated with gabapentin and duloxetine per os and with topical application of xylocaine, with signs of improvement after a three-month follow-up.

Case 4

A 47-year-patient, gravida 3, para 2, presented with a burning and painful sensation on the vulva with dyspareunia for the previous eight months. Clinically there were no findings of inflammation. The Q-tip test was positive, though. Many physicians proposed various examinations without setting a diagnosis. The patient was treated with fluoxetine per os and came for the follow-up visit after four months, showing slight improvement. She was to continue with the same regimen but she did not appear at her next follow-up visit.

Case 5

A 59-year-old patient, gravida 2, para 2, presented with intense pain on the vulva. She had suffered for three years and despite the various treatments from many different physicians,

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Table 1. — *Diagnostic procedures, disease duration, emotional status, treatment and outcome in all patients.*

Patients	Age	Number of medical specialities involved	Laboratory investigations	Symptom duration	Emotional disorders	Therapy	Outcome
G1	48	5	Blood, urine, vaginal excretion, biopsy	2 years	Neurotic & hypohondriac personality	Psychological support	Free of symptoms after one year and at a 5-year follow-up
2	52	4	Blood, urine, vaginal excretion, three biopsies	> 3 years	Signs of depression	Laser ablation	Stable at 10 months
3	38	4	Urine, vaginal excretion	3 months	Depression	Laser ablation Gabapentin, duloxetine	No response Improvement
4	47	4	Urine, vaginal excretion	8 months	None	Fluoxetine	Slight improvement after 4 months
5	59	4	Urine, vaginal excretion	3 years	None	Antidepressants, low calorie diet, anaplastic creams	Free of symptoms after one year
6	33	2	Urine, vaginal excretion, biopsy	15 months	None	Gabapentin	Improvement

there was a continuing deterioration of the burning sensation ending in her not tolerating her own undergarments. All investigations were negative. Application of topical steroids and topical anesthetics for a long period did not achieve relief of the symptoms. On clinical examination, which was extremely difficult due to the reaction of the patient to even a minor touch, there was mild erythema and slight atrophy of the minor labia mucosa and of the vulva. The patient reported an increase of her body weight of over 30 kg the last few years. After the long lasting application of steroids, and to avoid the risk of tachyphylaxis, only neutral anaplastic creams in combination with systemic anti-depressants and low calorie diet were proposed. At a follow-up visit one year later, the patient had lost twenty 25 kg of body weight and she was relieved from symptoms.

Case 6

A 33-year-old patient, gravida 0, para 0, presented for clinical evaluation as she had felt intense pain at the vulva and her clitoris for almost 15 months. These symptoms started after long treatment with econazole vaginal suppositories for a non established "candida vaginitis''. The patient reported dyspareunia and pain with tight clothing. She had been examined by a number of physicians and had undergone urine and vaginal excretion examination, as well as a biopsy, with no diagnostic findings. On clinical examination there was slight erythema of the vulva, probably due to the chronic application of topical xylocaine gel and topical steroids. The Q-tip test was positive. Treatment with gabapentin at a dose of 1200 mg per day for three months showed some efficacy at first but was discontinued by the patient herself due to persistent dizziness and sleepiness.

Discussion

For many decades the literature has hosted many different names for chronic vulvar pain (essential vulvodynia, dysesthetic vulvodynia, vulvar vestibulitis syndrome, vulvar dysesthesia, provoked vulvar dysesthesia, spontaneous vulvar dysesthesia, vestibulodynia, burning vulva, and clitorodynia). The classification of chronic vulvar pain to either vulvar pain related to a specific disorder or vulvodynia was proposed from the International Society for the Study of Vulvovaginal Disease in 2003 and offered a clear definition of terms [1].

Vulvodynia is today considered to be vulvar discomfort, most often described as burning pain, occurring in the absence of a relevant specific infectious, inflammatory, neoplastic or neurologic disorder. Localized or generalized, provoked or spontaneous, vulvodynia is rather a diagnosis of exclusion, reached commonly after a multidisciplinary approach and after a significant number of laboratory investigations. A great number of patients with vulvodynia seem to have a hidden or underecognized emotional instability. Vulvodynia has a severe impact on the patients' quality of life and its management remains a challenge.

According to the recently published evidence-based treatment guidelines, by the British Society for the Study of Vulval Diseases Guideline Group, an initial step of utmost importance is to explain the condition to the patient, allaying any fears and reassuring her that the condition is not infectious or related to cancer [2].

Initially, the importance of gentle genital hygiene should be emphasized, as well as elimination of excessive cleansing habits. Patients should also be encouraged to wash themselves with plain warm water and with a mild hypoallergenic and fragrance free nonsoap bar or cleanser, using hands only and not wash clothes or sponges [3].

From the topically applied agents, a trial of local anesthetics may be considered in all vulvodynia subsets. Systemically, tricyclic antidepressants (TCAs), e.g. amitriptyline or nortriptyline, are an appropriate initial treatment mainly for unprovoked vulvodynia. In addition to a TCA, gabapentin and pregabalin may be considered. Surgical excision of the vestibule or techniques to desensitize the pelvic floor muscles may be beneficial to a minority of patients with provoked pain. Selective serotonin reuptake inhibitors (SSRIs), like fluoxetine and serotonin norepinephrine reuptake inhibitors (SNRIs), such as venlafaxine and duloxetine, have also been used

to treat vulvodynia, but only in small trials. Intralesional steroid or botox injections may be considered in some cases [2, 3].

Few placebo-controlled studies have been conducted on medical treatments for vulvodynia. In the study by Bornstein *et al*., the true effectiveness of topical medications for the treatment of vulvodynia is questioned. The main finding of this study was that for low and high concentrations of topical nifedipine, as well as placebo ointment, mean pain intensity, assessed by the Q-tip test, speculum insertion and reports of sexual intercourse, was reduced at post-treatment compared with pre-treatment. What was really the benefit of the use of the topical medication? It was rather a placebo effect [4].

When psychological distress is expressed as a physical symptom and results in vulvodynia (somatoform hypothesis), then the necessity of treatment should be discussed. This vulvar pain may result in a conditioned avoidance response with spasm of the pelvic floor muscles. Depression and anxiety may contribute to a cycle of pain and patients may worry that sexual activity will be painful or vulvodynia may develop secondarily [5].

In our case series, all patients had undergone a number of laboratory examinations and many uneffective topical therapies (Table 1). When patients finally received a diagnosis of vulvodynia and were provided with clear and reliable information they expressed some relief. From the suggested regimens, only systemic antidepressants seemed to reduce the severity of symptoms. A possible explanation is that half of the patients had signs of depression. Was an underlying depression the cause of vulvodynia or did the vulvodynia cause anxiety and depression?

A crucial problem with vulvodynia patients remains the lack of awareness of this entity and the number of unnecessary investigations which lead to a delay in diagnosis and management. A recent survey demonstrated that there is lack of basic knowledge on chronic vulvar pain and vulvodynia among junior gynecologists who are being trained in tertiary units where most women with vulval pain conditions usually are referred to [6].

Conclusion

Vulvodynia remains a poorly recognized entity. Awareness and a multidisciplinary collaboration on vulvodynia are of great importance for early recognition and the appropriate management, as well as teaching seminars on this entity for all residents in dermatology and gynecology.

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