Vulvodynia associated with celiac disease


CASE REPORTS

Vulvodynia associated with coeliac disease Christine Costello Consultant (Haematology), J. R. Smith Senior Lecturer (Gynaecology) Chelsea and Westminster Hospital, London Case Report

A 44 year old woman presented with a six-month history of recurrent vulvovaginitis. She complained of burning, itching and a greenish vaginal discharge. The symptoms were worse before and after periods and soreness precluded the use of tampons. There was superficial dyspareunia with pain on penetration and splitting of the fourchette even when using lubricating jelly. General and abdominal examination was unremarkable. Genital examination revealed some atrophy of the external genitalia, inflammation and tenderness in the perivestibular area with Q-tip swab pressure. In addition a small ulcer in the anterior fornix of the vagina, a normal-looking cervix and a retroverted uterus with normal adnexae were found. Highvaginal and endocervical swabs were taken for culture and chlamydia detection and colonoscopy and vulvoscopy were performed. Vaginal pH was 5, and there was no evidence of bacterial, viral or chlamydia1 infection. Vaginal and cervical cytology were normal. Hormone assays did not suggest that the woman was menopausal. Colposcopy was normal, vulvoscopy revealed a small split at the posterior fourchette with no areas of aceto-white. Initial management was with advice to have saline baths, avoid perfumed soaps and avoid wearing tights. Dienoestrol@ cream (Ortho Pharmaceuticals Ltd, High Wycombe, Buckinghamshire, UK) was prescribed in view of the atrophic vagina although oestradiol levels were normal. The patient was empirically treated with oral metranidazole in view of the vaginal pH and observable discharge. Her partner was referred for genitourinary examination which proved to be normal. No improvement of the patient’s symptoms occurred, although the later addition of DaktocorP 1% cream (Janssen Pharmaceutical Ltd, Wantapp, Oxfordshire, UK) led to some improvement for a few weeks.

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Relapse then occurred with tenderness over the Bartholin’s glands and splitting at the fourchette and lateral to the clitoris. She was thereafter prescribed Demovatem cream (Glaxo Laboratories Ltd, Uxbridge, Middlesex, UK), Sensellem vaginal lubricant (LRC Products Ltd, Chingford, London, UK) and an oral anti-inflammatory agent. She improved substantially over the next two months but again relapsed. The patient was noted to have mild iron deficiency with haemoglobin 11.9 g/dL (12.0-16.0), mean corpuscular haemoglobin 26.4 pg (27-32), serum iron 5-1 pmol/L (10-30) and saturation 8.49% (20-55), but oral iron therapy failed either to improve her iron status or alleviate her symptoms. Further investigations were undertaken to elucidate the cause of the iron deficiency. Her periods were not heavy, and there were no symptoms of gastritis. She was slim but denied weight loss. She had felt excessively tired over the previous few months and had noticed abdominal bloating and excessive flatus. The bowels moved once daily and the stools appeared normal. The diet included meat, green vegetables and salads. B12 level was normal at 446 ng/L (160-800), but serum folate and red cell folate levels were reduced at 2 pg/L.
(3-15) and 85 pg/L (150-700). Serum ferritin (after several weeks of iron therapy) was reduced at 5 pg/L (8-140); serum calcium and serum proteins were normal. Antiendomysial IgA and antigliadin IgA antibodies were detected in the serum. These findings were very suggestive of malabsorption secondary to adult coeliac disease, a diagnosis confirmed on duodenal biopsy which showed a marked degree of villous atrophy and increase in chronic mononuclear inflammatory cells within the lamina propria and surface epithelium. Crypt hyperplasia was present. The patient began eating a gluten-free diet and took oral iron and folate supplements. Within a few weeks her vulval symptoms had markedly improved and by two months had completely resolved. Her energy level returned to normal and her symptoms of abdominal bloating and excessive flatus disappeared.

**Discussion**

Vulvodynia or vulvar vestibulitis is a recently described condition in which patients complain of chronic vulval discomfort with acute superficial tenderness and dyspareunia. The cause is probably multifactorial; infections with human papillomavirus and Candida albicans have been implicated in some patients, dermatological conditions such as lichen sclerosis and inflammatory dermatoses in others. Histopathological findings are consistent with achronic, nonspecific inflammatory response and occasionally metaplasia of the minor vestibular glands is seen. Standard gynaecological/genitourinary investigation of our patient with recurrent vulvovaginal discharge, soreness and vestibulitis revealed no evidence of bacterial or viral infection and no evidence of cytological abnormality. Empirical antibiotics, antifungal agents and anti-inflammatory therapy produced only temporary relief. Correction of her haematological deficiencies and management of her coeliac disease produced complete resolution of her symptoms.

Folates act as coenzymes in the transfer of single carbon units in reactions involved in DNA and RNA synthesis. Folate deficiency affects all rapidly growing (DNA synthesising) tissues and, in addition to its effect on the bone marrow (leading to megaloblastic erythropoiesis), folate deficiency also affects the epithelial surface of the mouth, stomach, small intestine and female genital tract, the cells showing macrocytosis with increased numbers of multinucleate and dying cells. Iron deficiency may also lead to mucosal changes but these are usually confined to the mouth and tongue. It is very probable that the fissuring noted in our patient was due to folate deficiency and the pruritus secondary to iron deficiency. The rapid response to treatment with folate and iron supplements along with a gluten-free diet suggests that her vulvodynia was associated with the effects of her coeliac disease. Iron treatment alone had been unsuccessful either in relieving the pruritus or in correcting the iron deficiency since she was not capable of absorbing iron adequately until gluten was excluded from her diet. We postulate that the cause of her vulvodynia was mucosal damage resulting from folate deficiency. It is very important that all concerned in the management of vulval disease are aware of the systemic causes of vulval symptoms.

**References**

This article, with no changes made in spelling or grammar, was found at http://www.celiac.com/gluten-free/topic/67690-vulvodynia-anyone/.