

Endometrioma of Bartholin's gland

Endometriom Bartholinovy žlázy

D. Habek¹⁻⁴, V. Kardum^{1,4}, J. Vlahović⁴

¹ Department of Gynecology and Obstetrics Clinical, Hospital Merkur Zagreb, Croatia

² School of Medicine, Catholic University of Croatia, Zagreb, Croatia

³ Croatian Academy of Medical Sciences, Zagreb, Croatia

⁴ School of Medicine, University of Zagreb, Croatia

Summary: We present a case of extrapelvic endometriosis of Bartholin's gland in a healthy 40-year-old woman with two deliveries. After the last delivery 10 years ago, she had cyclical pain with swelling in the Bartholin's gland area and dyspareunia with vulvodynia. Primary excision was performed, and Bartholin's gland endometriosis was pathohistologically proven as an extremely rare gynaecological entity. A follow-up examination 2 months after surgery proved complete recovery without vulvodynia and dyspareunia.

Key words: endometriosis – Bartholin's gland – vulvodynia

Souhrn: Prezentujeme případ extrapelvicke endometriózy Bartholinovy žlázy u zdravé 40leté ženy se dvěma porody. Po posledním porodu před 10 lety měla cyklické bolesti s otokem v oblasti Bartholinovy žlázy a dyspareunii s vulvodynií. Byla provedena primární excize a endometrióza Bartholinovy žlázy byla patohistologicky prokázána jako extrémně vzácné gynekologické onemocnění. Kontrolní vyšetření 2 měsíce po operaci prokázalo kompletní uzdravení bez vulvodynie a dyspareunie.

Klíčová slova: endometrióza – Bartholinova žláza – vulvodynie

Introduction

Endometriosis (endometrioma) of the Bartholin gland (EBG, glandula vestibularis major) is a very rare form of extrapelvic (extraperitoneal) endometriosis. The clinical picture and location of a tumor formation on the vulva can differentially indicate various tumor processes, most often a cyst of the Bartholin gland, but cyclic menstrual vulvodynia and dyspareunia can indicate the existence of possible endometriosis, primarily in the scar from an existing episiotomy or other perineal repair [1–3]. The implantation or metaplastic theory is the most common mechanism of EBG formation, and it is considered in a small number of publications. Transformation of the glandular epithelium of the Bartholin gland into endometriosis in cases without vulvovaginal repair has not been considered so far as a possible

scientific option for the origin of this rare pathological entity [2–4].

Only a few case reports of the EBG have been published in the literature, so considering the clinical importance and pandemic of endometriosis today, we present our case in this form.

Case report

A healthy 40-year-old woman **with two deliveries**, presented with a 10-year history of cyclic pain after her last delivery ten years ago, and swelling localized to the left Bartholin gland region, correlating with her menstrual cycle and progressively worsening for the past year with vulvodynia and dyspareunia. **Clinical inspection and palpation findings raised suspicion of scar endometriosis due to the patient's history of an episiotomy, as she reported localized pain in that area.** Examination revealed

a firm, painful tender nodule approximately 2 cm in diameter at the left region of the Bartholin gland which **was confirmed by ultrasound examination and a sharply limited hypoechoic avascular formation was found.** The lesion was non-erythematous and without signs of infection. Under intravenous anesthesia the entire suspected area was excised with primary suture reparation, revealing a 2 cm endometrioma located on the left side of the introitus with a typical cavity filled with chocolate-colored fluid (Fig. 1). The removed tissue was sent for histopathological analysis, which ultimately confirmed the diagnosis of endometrioma of the Bartholin gland. A control examination two months after the surgery proves complete recovery without vulvodynia and dyspareunia. **There is no information about the existence of**



Fig. 1. Left Bartholin gland endometrioma during surgical procedures.
Obr. 1. Vlevo endometriom Bartholinovy žlázy během chirurgických zákroků.

other sites of endometriosis, and apart from intermittent use of analgesics before surgery, the patient did not take any hormonal therapy before or after surgery.

Discussion

Although endometriosis primarily affects pelvic organs (intraperitoneal endometriosis), cases involving the Bartholin gland are still rare, which makes diagnosis challenging. In some cases, it can take up to 8–9 years to reach

a definitive diagnosis due to its nonspecific presentation and similarity to other gynecological conditions [1–5]. **Sošínik et al. analyzed 104 operated pathologies of Bartholin's glands and found retention cysts in 84.6% of cases, abscesses in 10.6%, extrauterine endometriosis in 2% and neoplasms in almost 3% of patients [6].**

The condition often mimics other gynecological disorders such as Bartholin cysts, abscesses, or neoplastic processes, delaying accurate identification

and treatment. Thus, almost all case reports indicated the primary diagnosis of a Bartholin's gland cyst, with vulvodynia in addition to premenstrual or perimenstrual cyclic pain, often in women with infertility, and after cystectomy the diagnosis of EBG is anecdotally found as an extremely rare entity [4,7]. Due to the cyclic nature of the symptoms characterized by pain and swelling that worsen during menstruation, clinicians should keep a strong level of suspicion for women experiencing persistent symptoms in the vulvar region. In this case, EBG is likely to have occurred during the last delivery and episiotomy, after which specific symptoms of cyclic vulvodynia appeared [1–7].

The pathogenesis of EBG is not completely understood, but theories suggest implantation during surgical procedures, delivery with obstetrical injuries or episiotomy, metaplasia, or lymphatic and vascular dissemination. **Maillard et al. recently published a meta-analysis of the diagnosis and treatment of cases of vulvoperineal endometriosis in 283 patients reports but mostly the implantation theory of this form of extraperitoneal endometriosis and the primary surgical excision procedure: in 95.3% presenting with vulvo-perineal endometriosis have undergone either episiotomy, perineal trauma or vaginal injury or surgery. Only 4.7% developed vulvo-vaginal endometriosis spontaneously i.e., without any apparent condition favoring it. Out of the 281 patients for whom a clinical examination was described, 97.5% patient showed a vulvo-perineal nodule, mass or swelling while 2.1% presented with bluish cutaneous lesions and 0.4% with bilateral polyps of the labia minora. All but one patients underwent surgical excision of their lesions but only 28.1% received additional hormonal therapy with recurrence rate was 10.2% [3].** The implantation theory during obstetric-surgical procedures is certainly the most commonly described; however,

there are cases where EBG appeared in infertile women without previous vulvar surgery, which supports the metaplastic theory [2,8]. Lavanderos et al. described a case of EBG in a patient with deep pelvic endometriosis, which supports the theory of metaplasia of the glandular epithelium of the Bartholini gland [1]. Gocmen et al. [9] showed a case of EBG in an infertile patient in whom laparoscopy did not prove the existence of intraperitoneal endometriosis.

Histopathological analysis remains the definitive confirmation of the diagnosis, and surgical excision is the primary treatment option, providing both symptomatic relief and definitive diagnosis. Hormonal therapies, such as GnRH analogs or oral contraceptives, have been explored in some cases, but their effectiveness in extrapelvic endometriosis remains uncertain [1–9].

Conclusions

Chronic cyclic vulvodynia and dyspareunia should arouse clinical attention to rare localization of endometriosis, such

as EBG, especially after previous deliveries with episiotomy or sutured vulvovaginal tears, and surgical excision with hormonal therapy in some cases is certainly a useful method of definitive therapy and helping to minimize discomfort in patients.

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ORCID of authors

D. Habek 0000-0002-7675-7064

V. Kardum 0000-0002-7397-344X

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prof. Dubravko Habek, MD, MSc, PhD

School of Medicine

Catholic University of Croatia

Ilica 244

10 000 Zagreb

Croatia

dhabek@unicath.hr

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