



Cervical endometriosis – a case report and review of literature

Cervikalna endometrioza – prikaz primera in pregled literature

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Abstract

Endometriosis is a disease in which endometrial glandular cells and stroma are present outside the uterine cavity. When the endometrial glands and stroma are located in the cervix, this is called cervical endometriosis. Clinically, cervical endometriosis most commonly presents as bloody vaginal discharge or extra cyclic bleeding. On cervical smears (CS), endometriosis may look like normal endometrial gland cells or atypical glandular cells of undetermined significance (AGUS), including adenocarcinoma in situ (AIS). We report a case of a 32-year-old woman who was admitted to the gynaecology department due to contact bleeding and a pathological colposcopy result.

Izveček

Endometrioza je bolezen, pri kateri so endometrijske žlezne celice in stroma prisotni zunaj maternične votline. Kadar se endometrijske žleze in stroma nahajajo v materničnem vratu, govorimo o cervikalni endometriozi. Klinično se cervikalna endometrioza najpogosteje kaže kot krvav vaginalni izcedek ali izvenciklična krvavitev. Na brisih materničnega vratu (BMV) lahko endometrioza izgleda, kot da bi šlo za normalne endometrijske žlezne celice ali kot atipične neopredeljene žlezne celice (AŽC-N), vključno z adenokarcinomom in situ (AIS). Članek predstavi primer 32-letne ženske, ki je bila sprejeta na oddelek za ginekologijo zaradi kontaktnih krvavitev in patološkega izvida kolposkopije.

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1 Introduction

Endometriosis is a benign disease, defined as the presence of endometrial glands and stroma outside the uterine cavity, frequently with associated bleeding and macrophages. The most commonly affected areas are the ovaries, uterine ligaments, recto- and vesicovaginal septum, peritoneum, pelvic cavity, umbilicus, inguinal and perianal regions; the cervix is less commonly affected (1). The lungs, brain and eyes can also be affected but rarely are (2). It is most common in women of childbearing age. The cause of endometriosis is explained by several theories. The in-situ theory advocates that endometriosis originates in extrauterine tissues, most commonly as a result of metaplastic changes, associated hormonal influences, inflammation or other biochemical or immunological factors; the transplantation theory is based on the concept of migration of normal endometrium («benign metastases») to extrauterine areas (3). Often, endometriosis is also associated with scars that occur after surgery on the uterus, including the cervix, fallopian tubes or after an episiotomy (1).

The most common symptoms and signs of endometriosis are dysmenorrhea, dyspareunia, pelvic pain, infertility and lower back pain that worsens during the menstrual cycle. In rare cases (<1%), malignant transformation of endometriosis may also occur (1,2,4).

Cervical endometriosis is present in 0.7–2.4% of patients (2,5,6). The prevalence of endometriosis is higher in patients with prior cervical procedures, such as a biopsy or conization (2,7). Vaginal delivery is also a risk factor for cervical endometriosis (8). Clinically, cervical endometriosis may be completely asymptomatic or present as bloody vaginal discharge and / or bleeding after sexual intercourse, less frequently as metrorrhagia or severe vaginal bleeding (5,7).

Medical history, various imaging methods (ultrasound – US, magnetic resonance imaging – MRI) and cytological examination of various types of samples (fine needle aspiration biopsy – FNAB, fluid cytology, cervical screening test) are all important for the diagnosis of endometriosis with histopathological examination being the final confirmation (9-12).

2 Case report

A 32-year-old patient was admitted to the Department of Gynaecology of our institution for diagnostic evaluation of extracyclic and contact bleeding and cervical erythroplakia. She had given birth three times, had

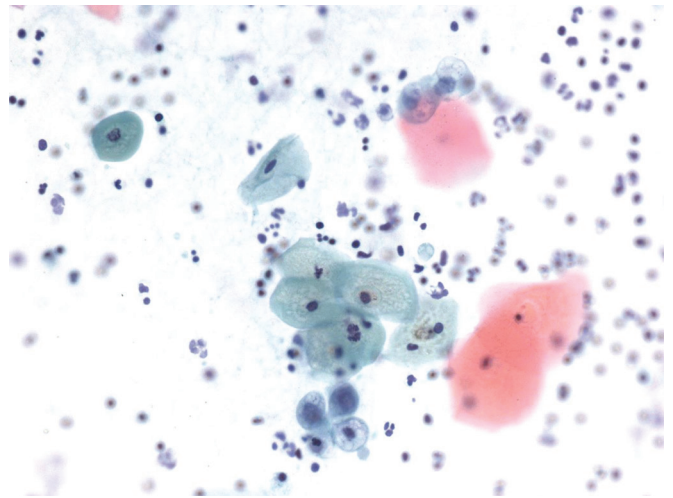


Figure 1: The last cervical screening test before re-LLETZ, Papanicolaou, 40x magnification.

had two miscarriages and regularly participated in the screening program for early detection of cervical cancer (ZORA). She was first invited for a cervical screening test in 2009; the result was normal. The first pathologic result was noted in 2012 as low-grade squamous intraepithelial lesion (LSIL). The control cervical screening test after one year was normal. After two years (2015) we again evaluated the cervical screening test as LSIL. The HPV test was positive. After surgery (large loop excision of the transformation zone - LLETZ), the histopathologic diagnosis was high-grade squamous intraepithelial lesion (HSIL; CIN3); the surgical margins were clear. All subsequent cervical screening tests were negative. At the last gynaecological examination in 2020 the patient reported contact bleeding; the gynaecologist described erythroplakia of the uterine portion of the cervix. A swab was taken and was negative. Due to contact and extracyclic bleeding the patient was referred to our institution, where an atypical transformation zone (ATZ) at 12 o'clock and erosions at 12, 7 and 5 o'clock were described on colposcopy. We repeated the cervical screening test, which was negative (Figure 1), however, based on the clinical presentation, we recommended an HPV test and follow-up according to accepted guidelines (13). Due to the previously described problems, we performed another cervical conization with an electric loop (re-LLETZ) and hysteroscopic endometrial biopsy.

We performed the histopathological analysis of the cervical cone sample, obtained with re-LLETZ, and the endometrial sample. The cone was by agreement marked at 12 o'clock. During the macroscopic examination, the samples were measured and described. The cone was

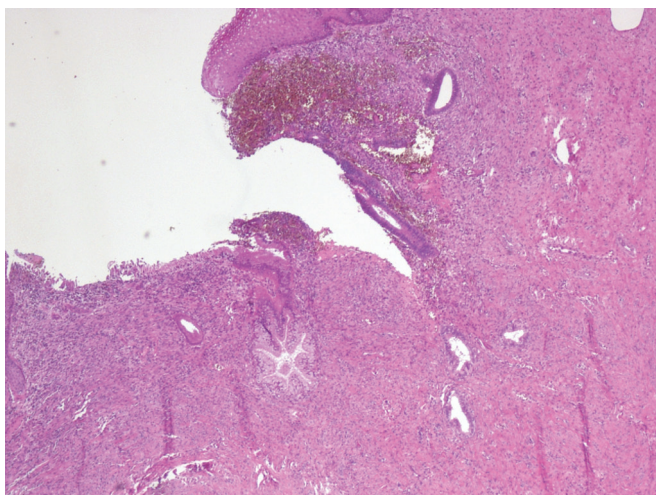


Figure 2: Focus of endometriosis with stromal bleeding in the transitional zone, HE, 10x magnification.

round in shape and measured 2.1x2.1 cm at the base. It was 1.1 cm tall from the base to the top. The external uterine orifice was positioned centrally. It was irregular in shape with the mucosa at the entrance of the orifice deeply wrinkled; it was smooth and shiny at the cervical portion. The cone was fixed in 10% neutral formalin. For histopathological examination, it was sampled in its entirety and cut into 7 consecutive tissue slices about 3–4 mm thick.

The endometrial biopsy specimen consisted of solid pink-black tissue samples up to 1.7 cm long and up to 0.5 cm in diameter. After pre-fixation in 10% neutral formalin, they were sampled in their entirety for histopathological examination.

After standard processing in a tissue processor, the slices were stained with haematoxylin-eosin.

With a light microscope, we found foci of glands with elongated nuclei surrounded by an endometrial stroma in the transitional zone in the cervical portion slices (Figure 2). There was bleeding in the stroma. The findings were consistent with a diagnosis of endometriosis in the cervical transition zone. A surface squamous epithelium ulceration and a scar after the previous procedure were also visible. The surgical margins were clear. The histological diagnosis of hysteroscopic biopsy of the uterine cavity was simple endometrial hyperplasia without atypia of the glandular epithelium.

3 Discussion

Cervical endometriosis is one of the rare types of endometrioses. In the literature, the prevalence is 0.7–2.4%, but no precise data are available for Slovenia (2,5,6). The most common problems reported by patients are

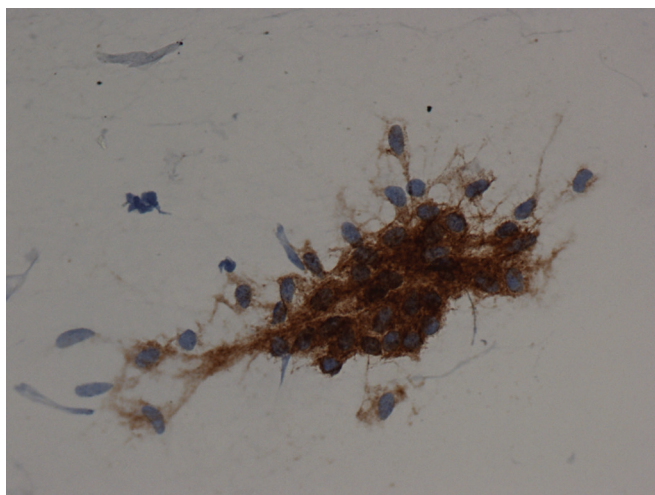


Figure 3: The last cervical screening test before re-LLETZ, positive CD10 reaction in stromal cells, 40x magnification.

bloody vaginal discharge and/or bleeding after sexual intercourse, less often metrorrhagia or severe vaginal bleeding. Deep cervical endometriosis may present with symptoms similar to endometriosis in general, such as dysmenorrhea, dyspareunia, pelvic pain, infertility, and lower back pain that worsens during the menstrual cycle (14). A serious but fortunately rare complication is extensive bleeding due to rupture of an endometrial cyst (15). Very frequently, however, endocervical endometriosis in particular can be completely asymptomatic. For the diagnosis of endometriosis, at least two of the three listed criteria must be met - the presence of endometrial cells, stromal cells and areas of bleeding with foamy macrophages (11). Although the histological diagnosis is normally simple, in rare cases only a stromal component may be present in the sample, mainly due to a poor sample (16). The diagnosis of cervical endometriosis based on a cervical screening test is frequently unreliable and can lead to misdiagnosis (14). With an incomplete history and absence of endometrial cells or stroma, the cervical screening test can be defined as normal. Sometimes, cervical endometriosis in a cervical screening test can be similar to tubal metaplasia, which is also a benign cervical lesion. An important criterion is the presence of cilia in glandular cells found in tubal metaplasia but not in endometriosis. Stromal cells usually occur in syncytial groups with oval and spindle nuclei and can be misidentified with elements of the lower uterine segment. With conventional cervical screening tests, additional staining can be difficult to perform due to only a single sample being taken. In our case, there were no clearly visible endometrial cells in the sample from the cervical screening test, but we did find some foamy macrophages (Figure 1). Following the diagnosis of cervical endometriosis,

the stromal component was identified retrospectively by cervical screening test sample decolorization and immunocytochemical staining for CD 10, which was positive (Figure 3).

An even greater danger is to overestimate the cervical screening test. The hyperchromatic crowded groups (HCG) with linear edges can be incorrectly identified as HSIL with endocervical gland invasion; an even more common differential diagnosis is glandular atypia of all grades, from atypical glandular cells, not otherwise specified (AGC-NOS) to severe atypical of high grade AGC, adenocarcinoma in situ (AIS) or even invasive adenocarcinoma (8,12,17). The reasons for this can be found in cytomorphological changes of glandular cells and stroma, which depend on hormonal fluctuations during the menstrual cycle (2,16). In glandular atypia, particularly severe, there is cell palisading, nuclear pseudostratification, rosette formation and feathering in the cervical screening test, possibly mitosis and apoptosis as well, and not only mild atypia or the presence of hyperchromatic crowded groups (6). In our experience, cytological diagnosis of endometriosis is easier and more reliable in other areas with a typical history and clinical presentation (pain and

increased changes related to the menstrual cycle).

4 Conclusion

Cervical endometriosis can present in a variety of ways, from a completely asymptomatic form to a form with severe vaginal bleeding. In cervical screening tests, the samples usually do not cover all morphological elements for a reliable cytological diagnosis of cervical endometriosis, and due to the small number of cases, we also do not have enough experience. Therefore, it is usually found subsequently after a known histopathological diagnosis. An accurate medical history, clinical presentation and information on previous operations are extremely important.

Conflict of interest

None declared.

Inform consent of the patient

The patient gave informed consent for the publication of her case.

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