CASE REPORT

DOI: 10.5336/caserep.2018-62453

A Rare Localization of Endometriosis: Labium Minus

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Received: 13.08.2018
Received in revised form: 03.10.2018
Accepted: 15.10.2018
Available online: 28.06.2019

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ABSTRACT Endometriosis on the labial localization is quite rare and there is only a few case reported in the literature. We report a patient complained of labial mass. Endemetriosis was not firstly estimated up to the operation when seen chocolate-colored mass content and the definite diagnosis was endometriosis in the labium minus according to histological pathology report. The endometriosis on the vulva may be suspected when cystic mass enlarging at menses. In this report, labial endometriosis will be discussed with clinical history and surgical approach.

Keywords: Endometriosis; vulvar disease; vulvar cyst

As known, Rokitansky was first described Endometriosis is the presence of the endometrial glands and stroma outside of the endometrium, pelvic or extra pelvic sites in 1860.[1] The endometrial gland and stroma can spread serviks, vagina, perineum or even spread urinary tract, gastrointestinal tract, thorax and nervous system.[2] The cases of extra-pelvic endometriosis are usually coincidental.[3] Because of unusual presentation, the exact diagnosis of endometriosis may resulting delay. We are reporting such an unusual case of endometriosis and surgical approach at this important sensory area hoping to increase awareness of this rare condition.

CASE REPORT

A 32-year-old female presented with a progressively enlarging labial mass over 9 months. She married for three years, with no medical and family history, nulligravida, with regular menstrual cycles, using oral contraceptive pills for contraseption. She reported that; the mass appeared to be more prominent and tender during menstrial period. She didnt complaint to dysmenorrhea, dyspareunia or chronic pelvic pain. There was no history of any operation, trauma and bowel or bladder complains. Pelvic examination revealed a 5cm x 4.5cm x 6cm, solitary and mobile cystic mass in her left labium minus (Figure 1a). Our preoperative diagnosis was a labial mucoid cyst. General physical examination and the transvaginal ultrasonographic (TVUS) examination were normal. There was no fluid or mass in the ab-

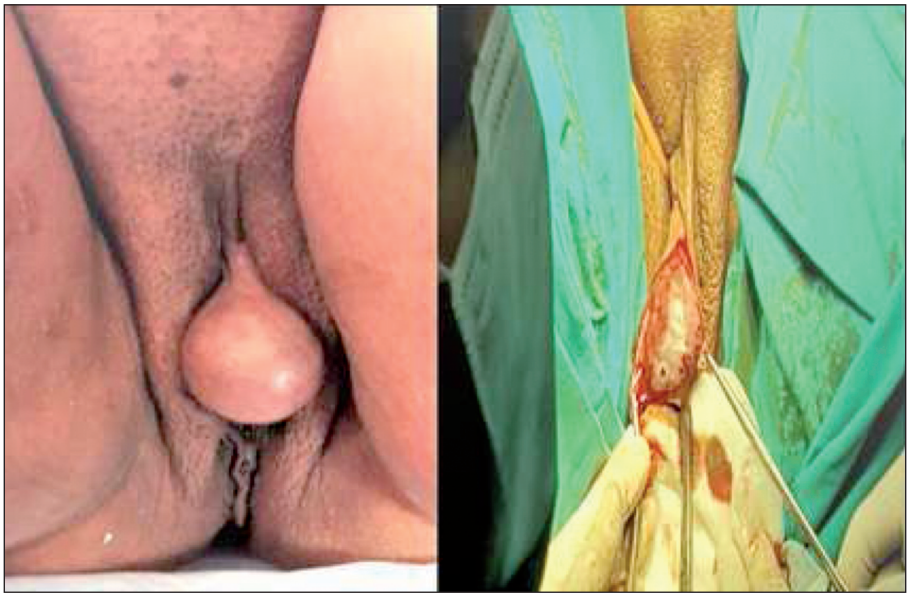

FIGURE 1: a) Preoperative image, **b)** Intraperative image.

dominal cavity and poush of douglass on TVUS. Perineal ultrasonography on vulva was performed and only fluid filled, regular edged cyst observed just under localized vulvar epitelium. There was no solid component and septa in the cyst. So, we didnt need Magnetic Resonance Imaging. Inspection of the vagina using a speculum as well as rectovaginal digital examination was uneven. Initially our diagnosis was a labial mucoid cyst and planned cystectomy under spinal anesthesia. Written informed consent and ethical approval were obtained from the patient for publication of this case report and taking images. Operation field was prepared with povidoine-iodine solution at dorsolithotomy position. The lidocaine with 1/1000 adrenaline infiltrated with injectable saline in order to hydrodissection. The incision was made on inter labial sulcus and during the operation, we encountered 5cm x4.5cm x 6cm, encapsulated, reguler edges cyst wall just under the subepithelial layer (Figure 1b). While dissection, the capsule was ruptured and a chocolate like fluid spread on the operation field. Totally the cyst capsule was removed with sharp and blunt disssections. The final diagnosis with pathology was endometrial epithelium, endometrial glands, and hemosiderin-laden macrophages present in labıal tissue. At the patient's 8-week postoperative evaluation, she had no recurrence cyst or tender at menses at this region. She reported no decreasing about her sexual arousal. The patient didnt receive extra treatment like Luteinizing Hormone Releasing Hormone (LHRH) analogues for postoperative good evaluation but she continued to take oral contraceptive pills for contraception.

DISCUSSION

Endometriosis is a immune-dependent genetically determined disease, which appears as an endometrioid tissue that grows outside the uterus affecting 1-7% of the population.[4] Clinically, endometriosis symptoms can be related both intra- and- extragenital organs. There is a case report about endometriosis of vulva and perineum in the literature.[5] Vulvar endometrial lesions usually occur after surgery or trauma.[6] However, spontaneously vulvar endometriosis reported only two cases in the literature.[7,8] The most common sites are episiotomy scars, Bartholin's gland and more rarely labia majora or labia minora.[8] Spontaneous vulvar location of endometriosis may be explained by the lymphovascular dissemination theory.[4] Extra pelvic

sites endometriomas are often confused with granuloma, hematoma, keloid, incisional hernia, vascular formation, sebaceous cyst, lipoma, or tumors and so patients often report general physicians, surgeons, or dermatologist because of atypical presentation and sometimes resulting delay in diagnosis.[9] Endometriosis at extra-pelvic sites often presents unusual symptoms. In our case, endometriosis may be suspected only due to the exacerbation of mass size during menses. Altough many of the cases are incidental, histology is landmark of the diagnosis. The accurate diagnosis of endometriosis involves the presence of two or more of these histologic results: endometrial epithelium, endometrial glands, endometrial stroma, and hemosiderin-laden macrophages.[10] Histological examination has also identified the labia minora as specialized, sexually responsive, highly vascular, folds of tissue with an abundance of neural elements.[11] Because of that, the surgeon should avoid unnecessary dissection of the labia minora. it makes sense to preserve important sensory areas during surgery. Preoperative sensory mapping of the labium and clitoral hood may prevent nociceptive injury, particularly in regions important in arousal pathways.[12] The typical midline incision risks disruption of the densely innervated clitoris and the labia minora. The sensory mapping technique leads to a lateral approach, minimizing this risk. Based on Wu et all's experience, in most instances, the safe zone falls at the junction of the labia majora and minora (the "inter-labial sulcus"), which is generally immediately before the transition to hairy skin.[13] We made the skin incision at interlabial sulcus and postoperative 8 months evoluation the patient reported no decreasing about her sexual arousal.

So, any vulvar lesion, regardless of its size, location and symptomps can be related to endometriosis. Surgical resection is a good approach to relieve the symptoms and provide histological proof. The surgeon should make sense to preserve important arousal area during the surgical resection.

Informed Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Source of Finance

During this study, no financial or spiritual support was received neither from any pharmaceutical company that has a direct connection with the research subject, nor from a company that provides or produces medical instruments and materials which may negatively affect the evaluation process of this study.

Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Design: *Adeviye Elçi Atılgan;* ***Data Collection and/or Processing:*** *Fatma Kılıç;* ***Analysis and/or Interpretation:*** *Fedi Ercan;* ***Article Writing:*** *Adeviye Elçi Atılgan.*

REFERENCES

1. Endometriosis. In: Fritz MA, Speroff L, eds. Clinical Gynecologic Endocrinology and Inferti-lity. 8th ed. Wolters Kluwer: Lippincott Williams & Wilkins; 2011. p.1221-48.
2. Acién P, Velasco I. Endometriosis: a disease that remains enigmatic. ISRN Obstet Gynecol. 2013;2013;242149. [Crossref] [PubMed] [PMC]
3. Singh KK, Lessells AM, Adam DJ, Jordan C, Miles WF, Macintyre IM, et al. Presentation of endometriosis to general surgeons: a 10-year experience. Br J Surg. 1995;82(10):1349-51. [Crossref] [PubMed]
4. Mahmood TA, Templeton A. The impact of treatment on the natural history of 106 endometriosis. Hum Reprod. 1990;5(8):965-70. [Crossref] [PubMed]
5. Binder SS. Endometriosis of vulva and perineum; report of a case. Pac Med Surg. 1965;73(5):294-6.
6. Odobasic A, Pasic A, Iljazovic-Latifagic E, Iljazovic-Latifagic E, Odobasic A, Idrizovic E, et al. Perineal endometriosis: a case report and review of the literature. Tech Coloproctol. 2010;14 Suppl 1:S25-7. [Crossref] [PubMed]
7. Nasu K, Okamoto M, Nishida M, Narahara H. Endometriosis of the perineum. J Obstet Gynaecol Res. 2013;39(5):1095-7. [Crossref] [PubMed]
8. Aydin Y, Atis A, Polat N. Bilateral endometrioma of Bartholin glands accompanying ovarian endometrioma. J Obstet Gynaecol. 2011;31(2):187-9. [Crossref] [PubMed]
9. Sengul I, Sengul D, Kahyaoglu S, Kahyaoglu I. Incisional endometriosis: a report of 3 cases. Can J Surg. 2009;52(5):444-5.
10. Medical Management of Endometriosis. ACOG Practice Bulletin #11; 1999.
11. Ginger VA, Cold CJ, Yang CC. Structure and innervation of the labia minora: more than minor skin folds. Female Pelvic Med Reconstr Surg. 2011;17(4):180-3. [Crossref] [PubMed]
12. Wagner IJ, Damitz LA, Carey E, Zolnoun D. Bilateral accessory breast tissue of the vulva: a case report introducing a novel labiaplasty technique. Ann Plast Surg. 2013;70(5):549-52. [Crossref] [PubMed] [PMC]
13. Wu C, Damitz L, Karrat KM, Mintz A, Zolnoun D. Clitoral epidermal inclusion cyst resection with intraoperative sensory mapping technique. Female Pelvic Med Reconstr Surg. 2016;22(3):e24-6. [Crossref] [PubMed] [PMC]