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Leiomyoma cellulare in the broad ligament of the uterus – case report and review of literature

Mięśniak komórkowy w więzadle szerokim macicy – opis sytuacji klinicznej i przegląd piśmiennictwa

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Abstract

Myomas are most often benign tumours of the female genital tract. Uterine fibroids are the most common myomas, while uterine cervix and intraligamentary ones are statistically less frequent. The most common histopathological form is the leiomyoma and the least common is the cellular leiomyoma (<5%).

Objectives: *As emphasized in the introduction, interlamellar parametrial cellular leiomyomas are extremely rare. Leiomyoma cellulare in the broad ligament of the uterus is the first case of that, therefore, the following situation deserves a short analysis and publication.*

Material and methods: *A 29-year-old patient was admitted to the Obstetrics and Gynaecology Clinic in Poznań due to a diagnosed lesion in the broad ligament of the uterus, possibly a myoma of the female genital tract. Cancer Antigen (CA) 125 level was 10.14 U/ml, beta sub-unit of human chorionic gonadotropin (β- HCG) was 0.1 mIU/ml, acute-phase protein was (CRP) 0.09 mg/l. The technique of laparoscopic myomectomy by enucleation of a 6-cm in diameter myoma was used after preparation of the right parametrium tissue. The patient left the hospital on the second postoperative day in good overall condition.*

Results: *Histopathology revealed leiomyoma cellulare oedematosum.*

Conclusions: *The above description of CL in the broad ligament of the uterus is a highly unique case and thus, deserves some attention, a short analysis.*

Key words: **myomas** / **fibroids** / **parametrium** / **leiomyoma cellulare** / / **the broad ligament** /

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Streszczenie

Mięśniaki są najczęstszymi łagodnymi guzami żeńskich narządów rozrodczych. Mięśniaki macicy rozpoznawalne są najczęściej podczas gdy mięśniaki szyjkowe i międzywięzadłowe występują statystycznie rzadziej. Najczęstszą formą histologiczną mięśniaków jest mięśniak gładkokomórkowy a najrzadziej rozpoznawalną (<5%) mięśniak komórkowy.

Cel pracy: *Jak wynika ze wstępu mięśniaki komórkowe międzyblaszkowe przymacicz występują bardzo rzadko, dlatego poniższy opis przypadku wart jest krótkiej analizy.*

Materiał i metody: *Pacjentka lat 29, została przyjęta do Ginekologiczno-Położniczego Szpitala Klinicznego w Poznaniu, z powodu zmiany w więzadle szerokim, prawdopodobnie o charakterze mięśniaka żeńskich narządów rodnych. Poziom antygenu nowotworowego CA 125 wynosił 10,14U/ml, podjednostki beta gonadotropiny kosmówkowej (β- HCG) 0,1mIU/ml, białka ostrej fazy (CRP) 0,09mg/l. W laparoskopii po odpreparowaniu tkanek okolicy przymacicza prawego, wyłuszczono mięśniak średnicy 6cm. Chora opuściła szpital w drugiej dobie po operacji, w stanie ogólnym dobrym.*

Na podstawie uzyskanego materiału tkankowego rozpoznano: leiomyoma cellulare (LC) oedematosum.

Wnioski: *Powyższy opis LC w więzadle szerokim jest jedynym takim przypadkiem, dlatego wart jest krótkiej analizy.*

Słowa kluczowe: **mięśniaki** / **przymacicza** / **mięśniak komórkowy** / **więzadło szerokie** /

Introduction

Myomas are most often benign, monoclonal, hormone-dependant tumours of the female genital tract, composed of smooth muscle cells and fibrous connective tissue [1, 2]. Statistically, they are often found in women between 40-50 years of age (most imaging techniques lack resolution and thus only 12% of patients under 35 years of age may be diagnosed), nulliparas, obese, Afro-American women and their next of kin, as well as in cases of multiple cutaneous and uterine leiomyomatosis [1, 2, 3, 4]. The growth of myomas seems to be related to hormonal imbalance, as they are not present before pubescence, less frequent after menopause or after pharmacological and surgical gonadectomy [2]. Also, bone morphogenetic protein 1 enzymes (BMP-1/Muld), HMGA 2 and HMGA 1 genes, growyh hormones, cytokines and, possibly, other yet unidentified factors, may play an important role n the uncontrolled cell growth of tissue with extracellular matrix [1, 2, 5]. Regardless of the fact that the tumour may be asymptomatic, on average in 20-50% of cases it may cause menorrhagia, pelvic pain, dysfunctions of the urinary tract and obstructive defecation, infertility, spontaneous abortion, premature labour [6].

In the case of patients with myomas, bleeding is most often manifested as heavy menstrual bleeding (menorrhagia), which is influenced by the location and size of the myoma [7]. Depending on the location, the following myomas may be distinguished: uterine myomas (subserosal, intramural, submucosal) and uterine cervix myomas, peduncular or intraligamentary (interlamellar) [2]. Uterine fibroids are the most common myomas, while uterine cervix or intraligamentary ones are statistically less frequent [2]. On the other hand, regardless of their primary location but depending on their histopathological structure, fibroids may be divided into: smooth cell, epithelial, peculiar, liposarcoma and cellular leiomyomas CL (leiomyoma cellulare). The most common histopathological form is the leiomyoma and the least common is the cellular leiomyoma (<5%). Cellular leiomyomas contain more fibres of smooth muscles than smooth cell leiomyomas and are defined by the Word Health Organization as typical leiomyomas that exhibit hypercellularity [8, 9].

Due to its form, cellular leiomyoma is often suspected to be a malignant hyperplasia, particularly if the mitotic index is ≥5 and is accompanied by cellular atypia [2, 5, 10].

The risk of a cellular leiomyoma undergoing malignant transformation into a smooth cell sarcoma is low (0.1–0.8%) [2].

As emphasized in the introduction, interlamellar parametrial cellular leiomyomas are extremely rare. Leiomyoma cellulare in the broad ligament of the uterus is the first case of that, therefore, the following situation deserves a short analysis.

Case report

A 29-year-old patient was admitted to the Obstetrics and Gynaecology Clinic in Poznań due to a parametrial lesion, possibly a myoma of the female genital tract. Gynaecological history: first menstruation at the age of 12, regular bleeding, a 26-day cycle. The patient had one spontaneous abortion at 6 weeks gestation, four years prior to the admission. General medical history: asthma, no medication. Non-smoker, did not use contraceptives. Gynaecological examination revealed antecurvature of the corpus uteri, movable, pain-free, uterine cervix with closed external orifice, normal adnexa on both sides, no pathological changes, in the right parametrium there was a visible, 6-cm in diameter resistance, discharge was normal.

Vaginal probe ultrasound examination of the organs of the lesser pelvis revealed antecurvature of the corpus uteri, regular size and shape. Endometrium thickness: 6mm. In the right parametrium a 6-cm lesion was discovered, possibly a uterine myoma. Both ovaries were normal, with small follicles. The Pouch of Douglas was normal. Cancer Antigen (CA) 125 level was 10.14 U/ml, beta sub-unit of human chorionic gonadotropin (β-HCG) was 0.1mIU/ml, acute-phase protein (CRP) was 0.09mg/l. The remaining tests (among others: morphology, electrolytes, general urine test) proved to be normal as well. After gynaecological examination and lab tests results, the decision to perform laparoscopic myomectomy to remove the uterine fibroid was made. Under general anesthesia the tissues of the right parametrium was prepared and a 6-cm in diameter myoma was enucleated. (Figure 1).

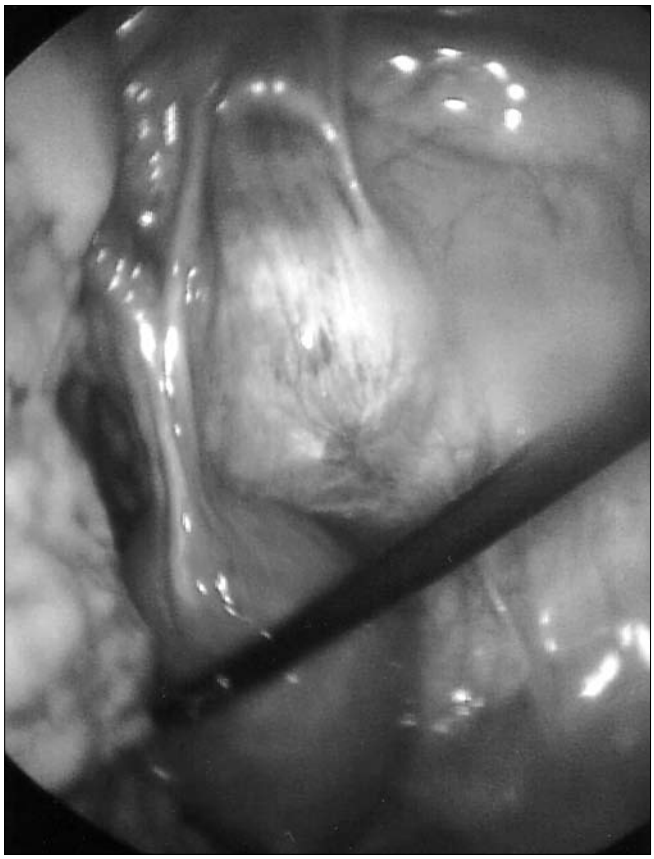

Figure 1. Fibroid in the right broad ligament of the uterus.

The course of the surgery and post-operative recovery period were uneventful. The patient was discharged on the second postoperative day in good overall condition. Histopathology revealed the myoma to be leiomyoma cellulare oedematosum.

Discussion

Cellular myomas, due to their origin from smooth muscle tissue, may be located in almost any organ. Uterine cellular myomas are the most common, accounting for about 95% of all benign lesions of the genital tract. Descriptions of intraligamentary (interlamellar) location of the fibroid can rarely be found in literature. These are usually case studies, single cases of diagnosis of broad or round ligament fibroid. The diagnosis of myomas is based on medical history, gynaecological and ultrasound examinations. Ultimately, a pathomorphologist confirms the diagnosis on the basis of microscopic evaluation. Differentiation between cellular myoma and leiomyoma presents some diagnostic challenges. In a magnetic resonance imaging test, Schwarz et al., [11] noted that cellular leiomyomatosis is uniformly of slightly greater signal intensity to the normal myometrium but hypointense to surrounding pelvic fat on T1-weighted images [11]. The question arises whether it is indeed possible to diagnose CL on the basis of medical history or individual features of a patient. Taran et al., [10] analyzed retrospectively histopathologic results and medical histories of patients operated on due to uterine myomas between January 1989 and December 2008.

The study group included 99 women with diagnosed cellular myoma and the control group comprised 198 patients with diagnosed uterine leiomyomas. The weight of the myoma and the uterus itself was greater among the women with CL cases compared to women with typical leiomyomas (OR 2.1, 95% CI 1.3-3.4). Additionally, cellular leiomyomas were diagnosed more frequently in younger women complaining of greater pain and those with coexistent adenomyosis and endometriosis. In multivariable logistic regression analyses, women with CL were more likely to have surgical indication for enlarging leiomyoma (OR 7.1, CI 2.4-25.1), were more likely to have more fibroid burden (OR per doubling in fibroid size 1.2, CI 1.1-1.3) and have fewer leiomyomas (OR 0.9, CI 0.9-1.0) when compared to women with typical leiomyomas. There were single cases of LC but they proved to be more solid in the histopathological examination. Hence the conclusion that cellular leiomyomas are rather rare and in the histopathological examination they reveal features of not only myomas but also of leiomyosarcomas [10].

Regardless of the considerable development of ultrasonography, differentiation between leiomyomas and leiomyosarcomas remains challenging. Hypoechogenic areas typical of leiomyosarcomas may also be found in necrotic myomas, whereas rapid tumour growth, features of neovascularity, significant flow-rate and low resistance flow are characteristic for malignant lesions [5]. Immunohistochemical tests, for example desmina, CD 10, h-caldesmon expression, seem to be useful in the differentiation [5]. On the other hand, Banaczek et al., [5] performed a detailed analysis of 309 surgeries due to uterine fibroids between January 2000 and June 2002. Cellular leiomyomas were diagnosed, based on the microscopic evaluation, in only 4.53% of women (n=14), aged 44.5±11.15 years (from 29 to 73 years of age) and the age was lower than in the control group. These were mostly single tumours, 7.54cm in diameter (5 to 11cm). Nevertheless, clinical symptoms, USG imaging and macroscopic LC were characteristic for leiomyomas. In the authors' opinion, because of microscopic similarities to other stromic tumours of the body of the uterus and myo-levicellular sarcoma it is necessary to examine a larger number of specimens and to use valid criteria of recognition [5].

Most patients are asymptomatic and are diagnosed incidentally, but among symptomatic patients pelvic pain, palpable mass or abnormal bleeding are the most common symptoms similar to those caused by leiomyomas of the broad ligament. Salman et all. [12] reported a sixth case of lipoleiomyoma of broad ligament which was diagnosed in a postmenopausal woman who underwent exploratory laparotomy with a preoperative diagnosis of a solid adnexal mass suggesting an ovarian malignancy.

In the present case described by the authors, the patient was an asymptomatic and relatively young person, nullipara, but with obstetric history. Moreover, she had not used contraceptives and the USG image was typical for leiomyoma, except that it was located in the right parametrium. Information and research in the generally available medical databases regarding leiomyomas and/or interlamellar fibroids is scarce and there are only single cases.

Conclusions

The presented example of CL in the broad ligament of the uterus is the only such case and, thus, particularly worthy of analysis.

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