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Ovarian leiomyoma – case reports and literature review

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SUMMARY

Introduction. Ovarian leiomyoma is a rare benign ovarian tumor accounting for 0.5–1.0% of all benign ovarian tumors. The paper presents four cases of ovarian leiomyoma detected post-operatively by histopathological examination in patients treated at the Gynecology and Obstetrics Ward of the Specialist Hospital in Radom, Poland, in the period of 1998–2017.

Material and methods. A retrospective analysis involved surgical and histopathological reports. A 51-year-old patient underwent hysterectomy with bilateral removal of the adnexa. Three patients had the uterine body amputated: in a 49-year-old patient, the adnexa were preserved and a myoma of the right ovary was excised; in a 59-year-old patient, the adnexa were removed bilaterally and a myoma of the rectouterine pouch was excised; in a 54-year-old patient, the adnexa were removed bilaterally. Final diagnoses were made on the basis of post-operative microscopic examination of the specimens. Moreover, the article reviews accessible literature on ovarian leiomyoma.

Results. Leiomyomas were located on the right side in all cases. Concomitant occurrence of endometriosis and ovarian leiomyoma was detected in two cases, and in one of them uterine myomas were accompanied by a myoma in the rectouterine pouch.

Conclusions. A diagnosis of ovarian leiomyoma is made on the basis of a histopathological examination. Surgical treatment depends on patient's age, her intention to remain fertile, the extent of the lesion and concomitant problems affecting her genital organs.

Key words: ovarian leiomyoma; rare ovarian tumors

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INTRODUCTION

Ovarian leiomyoma accounts about 0.5–1% of all benign ovarian tumors [1,2]. It is a mesenchymal tumor deriving from the smooth muscle tissue. Ovarian leiomyoma cells can originate from vascular smooth muscles as well as from smooth muscle of the ovarian ligaments, multipotent cells of the ovarian stroma or undifferentiated germ cells [3–5]. According to Guanasekaran et al., approximately 80 cases of these tumors have been reported in the literature so far [6].

Ovarian leiomyoma is usually detected incidentally during surgery or post-mortem examination [7,8]. The final diagnosis can be made after a histopathological examination [1,4,9–12]. On histopathology, there is no nuclear atypia, nuclear pleomorphism or mitotic activity. The nuclei express positive staining for estrogen receptor (ER) and progesterone receptor (PR), which attests to estrogen and progesterone impact on leiomyoma growth [7]. Differential diagnosis includes fibroma/thecoma-type tumors and leiomyosarcoma [1,13]. Immunohistochemical staining, e.g. desmine, α-inhibin, calretinin and smooth muscle α-actin (α-SMA), is useful and used in histopathological and differential diagnosis [7,8,14,15].

In symptomatic cases, patients present with abdominal pain, vomiting, sometimes hydronephrosis, Meigs' syndrome, elevated Ca-125 value and ascites with concomitant vulvar edema (atypical Meigs' syndrome) [16,17]. Disorders of menstruation and fertility are common [4,18]. In most cases, ovarian leiomyoma develops in premenopausal (80%) and postmenopausal women, usually unilaterally. These tumors tend to be rather small, with diameters not exceeding 3 cm [1,8,10,11]. Bilateral occurrence and large size have been noted in girls and young women at child-bearing age [2,10,11]. Very large cystic ovarian leiomyomas show signs of degeneration: cystic degeneration, myxoid degeneration with multiple foci of hemorrhage, thrombosis and necrosis [7]. A case of

torsion of the right ovary with a leiomyoma has been reported by Bhageerathy et al. [19]. The co-occurrence of ovarian leiomyoma with uterine fibroids was noted by Lerwill et al. [13] in 54.6% of cases and Doss et al. in 78% of cases [1]. The report on lipoleiomyoma of the uterus with concomitant ovarian leiomyoma is the only case in the literature [20]. In one case, authors present bilateral ovarian leiomyoma concomitant with ovarian cancer [5]. Also, other pathological conditions of the ovary have been noted to co-occur with ovarian leiomyoma, either on the ipsilateral or contralateral side [21,22,23]. The histological diversification of smooth muscle tumors in the ovary is similar to that in the uterus [1,13].

AIM

The aim of the paper was to present four cases of ovarian leiomyoma and review available literature on this rare tumor.

MATERIAL AND METHODS

The retrospective analysis involved medical records of patients operated in 1998–2017 in the Gynecology and Obstetrics Ward of the Specialist Hospital in Radom. In this period, histopathological examinations revealed four cases of ovarian leiomyomas, all on the right side. Patients were selected for surgery based on a pelvic examination, ultrasound scan and histological assessment of uterine scrapings. Ultimate diagnoses were made in the Department of Pathomorphology of the Specialist Hospital in Radom. Moreover, the accessible literature on ovarian leiomyoma has been reviewed.

CASE PRESENTATIONS

Case 1

A 51-year-old patient was admitted to the Gynecology and Obstetrics Ward on December 7 1998 for surgery due to uterine fibroids. A month before, she had undergone diagnostic uterus abrasion. The histopathological examination revealed: *endometrium in stadio secretionis*. The patient said that the menarche had appeared at the age of 13 years. Recently, menstruations were irregular, every 28–32 days, prolonging to 6 days and painful.

On admission, her state was good. Body mass was 65.0 kg, and body mass index was 23.88. RR: 140/80 mm Hg, heart rate: 78/min, temperature: 36.6°C. Laboratory parameters were within normal ranges. A pelvic speculum examination demonstrated as follows: cylindrical, well-developed vaginal portion; oval, shut external opening; the vaginal portion of the cervix clear of pathological changes; mucoserous physiological discharge. The bimanual examination revealed enlarged uterine body: 180 x 120 mm. The adnexa were impalpable bilaterally, the parameteria were normal. An ultrasound examination showed the uterine body of heterogeneous echotexture with numerous hyperechoic lesions, probably fibroids, with the size of 200 x 150 x 80 mm and the heterogeneous endometrium with the thickness of 12 mm. The structure of the left ovary, measuring 29 x 20 mm, was normal. On the right side, the ovary with a hyperechoic lesion measuring 30 mm was seen.

The patient was deemed eligible for laparotomy from the inferior midline incision. The enlarged uterine body was confirmed (200 x 150 x 80 mm). There were no macroscopic lesions in the adnexa. The remaining abdominal organs were normal on macroscopic inspection and palpation. The patient underwent hysterectomy with bilateral salpingo-oophorectomy, performed in the typical manner. She was discharged on day eight after the procedure. The histopathological examination revealed: 1) body of the uterus: *Leiomyomata intramuralia partim hyalinisantia et endometriosis superficialis*; 2) right ovary: *Leiomyoma hyalinisans* (dimensions: 10 x 5 x 5 mm.)

Case 2

A 49-year-old patient was hospitalized at the Gynecology and Obstetrics Ward for surgery due to uterine fibroids. Two months before, she had undergone diagnostic uterus abrasion. The histopathological examination revealed: *Hyperplasia endometrii simplex*.

The patient said that the menarche had appeared at the age of 14 years. Recently, menstruations were prolonging to 7 days and painful. She had delivered 2 children, both naturally. She reported one spontaneous miscarriage. On admission, her state was good. Body mass was 64.0 kg, body mass index 26.30, RR: 140/90 mm Hg, heart rate: 78/min and temperature: 36.6°C. Laboratory parameters were within normal ranges. A pelvic speculum examination demonstrated as follows: cylindrical, well-developed vaginal portion; slit-like, shut external opening; vaginal portion of the cervix clear of pathological changes; mucoserous phy-

siological discharge. The bimanual examination revealed enlarged uterine body: 120 x 100 mm. The adnexa were impalpable bilaterally, the parameteria were normal. An ultrasound examination showed the anteflexed uterus with the size of 117 x 116 x 90 mm, heterogeneous echotexture with numerous hyperechoic lesions, probably fibroids, the largest of which reached 70 mm in diameter, and the endometrium with clear margins and the thickness of 11 mm. The left ovary measured 29 x 20 mm and was normal. The right adnexa: right ovary measuring 30 x 20 cm with a 10 mm hyperechoic lesion.

The patient was deemed eligible for laparotomy from the transverse Pfannenstiel incision. The enlarged tumorous uterine body was confirmed (130 x 100 x 90 mm). The tumor in the right ovary was confirmed as well (10 mm in diameter). Apart from this, there were no macroscopic lesions in the adnexa.

The uterus was removed without the adnexa and the ovarian tumor was excised. The postoperative period was uncomplicated. The patient was discharged on day 4 after surgery. Histopathology revealed: 1) Body of the uterus: *Leiomyomata intramuralia*. *Endometriosis*; 2) a fragment of the right ovary: *Leiomyoma parvum hyalinisans*.

Case 3

A 59-year-old patient was admitted for surgery due to uterine fibroids and right ovarian tumor. Diagnostic abrasion showed *Polypus canalis cervicalis*, *Endometrium atrophicum*. The patient said that the menarche had appeared at the age of 13 years. Menstruations were regular, every 28 days. The last one occurred at the age of 56 years. The patient had never given birth or miscarried. She was operated for uterine fibroid at the age of 33 (myomectomy).

The patient complained about pain in the low abdomen and lumbosacral region. She was treated for arterial hypertension and type 2 diabetes mellitus. Laboratory findings were insignificant. A pelvic speculum examination demonstrated as follows: cylindrical well-developed vaginal portion; oval, shut external opening; vaginal portion of the cervix clear of pathological changes; mucoserous physiological discharge. The bimanual examination revealed enlarged uterine body: 120 x 100 mm, shifted to the left. The evaluation of the adnexa was difficult because of a tumor located over and behind the uterus on the right side in the pelvis minor and mid-abdomen, measuring 100 x 90 mm. An ultrasound examination showed enlarged uterine body with the size of 110 x 90 x 70 mm, with heterogeneous echotexture and numerous hyperechoic lesions, probably fibroids, the largest of which reached 70 mm. The endometrium was heterogeneous with the thickness of 11 mm. Over and behind the uterus, there was a hyperechoic lesion measuring 110 x 100 x 70 mm. The adnexa were impalpable.

The patient was deemed eligible for laparotomy from the inferior midline incision. The enlarged uterine body with numerous intramural fibroids and left-lateral shift was confirmed. The posterior uterine wall was in solid adhesion with the sigmoid colon. The right ovary with the tumor measured 120 x 80 x 75 mm. There was a 50 mm tumor in the rectovaginal pouch. The adnexa were excised bilaterally with the right ovarian tumor. Upon releasing adhesions, the leiomyoma was dissected from the rectovaginal pouch, and then the uterine body was removed. The postoperative period was uneventful. The patient was discharged on day 5 after surgery. Histopathology revealed: 1) body of the uterus: *Leiomyomata multiplicia*; 2) the right adnexa (the right ovary in the form of an encapsulated tumor measuring 120 x 80 x 75 mm and of band-like structure in the cross section; a fragment of the ovary with the cyst) – *Leiomyoma, Ovarium cum cystis endometriotica;3)* tumor of the rectovaginal pouch: Leiomyoma.

Case 4

A 54-year-old patient was admitted for surgery due to uterine fibroids and right ovarian tumor. Diagnostic uterus abrasion revealed *Mucosa colli uteri*. Single bands of atrophic glandular epithelium of the uterine body.

The patient said that the menarche had appeared at the age of 14 years. Menstruations were regular, every 28 days. The last one occurred at the age of 52 years. She had delivered 3 children, all naturally. No miscarriages. The patient was treated due to arrhythmia and arterial hypertension. Laboratory findings were insignificant. A pelvic speculum examination demonstrated as follows: cylindrical, well-developed vaginal portion; slit-like, shut external opening; vaginal portion of the cervix clear of pathological changes; mucoserous physiological discharge. The bimanual examination revealed slightly enlarged uterine body in the interme-

diate position: 60 x 50 mm. It was movable and not painful on palpation. A flexible tumor with the diameter of 60 mm was found in the region of the right adnexa. The left adnexa were impalpable.

Ultrasonography revealed the uterine body in an intermediate position of heterogeneous echogenicity and the size of 60 x 50 x 40 mm. The endometrial thickness was 3 mm. A hyperechoic 6.22 mm lesion was noticed in the uterine cavity, probably an endometrial polyp. The right adnexa: the right ovary with a hypoechoic lesion of dense contents and the size of 80 x 60 x 50 mm. The left ovary, measuring 29 x 15 x 10 mm, was normal.

The patient was deemed eligible for laparotomy from the transverse Pfannenstiel incision. The body of the uterus was found to be slightly enlarged, with a 20 mm intramural fibroid in its anterior wall. The right ovary, with the cyst measuring 100 x 80 x 50 mm and a solid tumor next to the cyst measuring 20 mm, was identified. The adnexa were excised and sent for a histopathological evaluation. After the result had arrived (a benign lesion), the uterine body and the left adnexa were removed in the typical manner. Histopathology revealed: 1) the right adnexa: ovary: a cyst measuring 100 x 80 x 50 mm with distended fallopian tube, filled with clear, stretchy fluid and smooth lining. Next to the cyst, there was a 20 mm tumor, off-white, band-like, solid and shiny in the cross section: *Cystis mucinosa ovarii.Leiomyoma*; 2) uterine body (measuring 60 x 50 x 35 mm) – *Leiomyomata intramuralia corporis uteri.Polypus endometrii.*

The postoperative period was uneventful. The patient was discharged on day 4 after surgery.

DISCUSSION

In most cases, ovarian leiomyoma develops unilaterally, usually before or after menopause [10,11]. However, such diagnoses have been made and surgical treatments conducted even in patients aged 17 and 79 years [11,15]. Ovarian leiomyoma is frequently concomitant with uterine fibroids [1,11]. It does not occur in children [10].

In our material, the co-occurrence with uterine fibroids was noted in two cases, including one with another accompanying leiomyoma of the rectovaginal pouch. Also in two cases, leiomyomas were concomitant with endometriosis. *Leiomyoma ovarii* can be a benign metastasizing leiomyoma (according to the WHO classification from 2003 – 8898/1).

In the case of fibroid uterus, fibroids can be additionally supplied by the ovarian vessels, which makes metastases possible. Various MRI techniques may be helpful in diagnosing these lesions. However, due to high costs, such examinations are rarely ordered for routine assessment of uterine fibroids [24]. The development of leiomyomas in the organs of the pelvis minor, abdominal or peritoneal cavities and their co-existence with uterine fibroids are also explained with a hypothesis of parasitic leiomyomas. This situation can take place in the case of detachment of a subserous myoma [25]. Ovarian leiomyomas are linked with other ovarian pathologies, either ipsilateral or contralateral, or very rarely bilateral. These include reported cases of: *cystis endometriotica* (in the same ovary) [21], *cystadenofibroma serosum* (in the same ovary), which has been presented as the first report of these tumors occurring together [23], and *cystadenoma mucinosum* (in the same ovary) [9]. The co-existence of an endometrioid cyst was noted in the third and serous cyst in the fourth presented case. Of various histological ovarian leiomyoma subtypes, cellular, mitotically active, atypical and mucosal leiomyomas have been noted in the literature. Of malignancies, leiomyosarcoma has been described [1,13].

Laboratory findings in patients with ovarian leiomyoma include cases with high CA-125 values and a case with high inhibin B and luteinizing hormone (LH) levels [18,26,27]. In all patients with Meigs' syndrome or atypical Meigs' syndrome, ascites and pleural effusion subsided after surgical treatment [9,21,26–28].

Because of its rare occurrence, ovarian leiomyoma is diagnosed only during intra- and/or postoperative examinations [29]. Koo et al. report that all nine patients with postoperative diagnosis of ovarian leiomyoma were misdiagnosed preoperatively (leiomyoma was diagnosed as pedunculated leiomyoma, fibroma or ovarian endometriosis) [4]. Undoubtedly, this is also associated with the fact that these lesions are in most cases relatively small. The four cases of ovarian leiomyoma presented above were also diagnosed in postoperative histopathological examinations.

It is difficult to distinguish ovarian leiomyoma from other ovarian tumors in ultrasound or computed tomography [8]. Other authors report that preoperative diagnosis, even using computed tomography (CT) and magnetic re-

sonance imaging (MRI), has led to the identification of ovarian serous cystadenoma, fibroma and/or thecoma [9,27,28,30].

Transvaginal US shows ovarian leiomyoma as a homogeneous isoechoic mass with no pathological flow on Doppler US. MRI visualizes it as a solid tumor with low-intensity signals in both T1- and T2-weighted images [12].

Treatment of this tumor in perimenopausal women or in women with concomitant uterine conditions usually involves subtotal or total hysterectomy with uni- or bilateral removal of the adnexa or only with ovarian leiomyoma excision. Fertility-sparing procedures are conducted in girls and women who wish to remain fertile [8,19].

CONCLUSIONS

A diagnosis of ovarian leiomyoma is made after a histopathological examination. Surgical treatment depends on patient's age, her intention to remain fertile, the extent of the lesion and concomitant problems affecting her genital organs.

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