Case Report

Pelvic Retroperitoneal Cellular Leiomyoma: A Case Report

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Leiomyomas are common benign gynecological tumors and usually arise in the uterus. The retroperitoneal cellular leiomyoma, one of the unusual manifestations, is a rare tumor. Diagnosis and treatment are challenges. We report a case of 65-year-old women presented with an asymptomatic mass beneath the right posterior vaginal mucosa. CT imaging revealed heterogeneous mass 6 cm in the pelvic cavity abutted lower segment of uterus, cervix, and vagina. The provisional diagnosis was subserosal cervical leiomyoma. She underwent exploratory laparotomy. Intra-operative, a normal size uterus was found separately from retroperitoneal pelvic mass at the level of internal os. Histological report confirmed cellular leiomyoma later. Total hysterectomy, bilateral salpingoophorectomy and completely excision of tumor were achieved with good outcome. Our patient represents the rare case of retroperitoneal cellular leiomyoma, which is hardly identified from internal examination and preoperative imaging. Surgical removal is essential for pathological diagnosis and treatment.

Keywords: *Leiomyoma, Retroperitoneum, Cellular leiomyoma, Vaginal mass*

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Leiomyoma is the most common tumor of the uterus in female pelvis. They are responsible about one third of all hospital admission to gynecological services. It affects 20-30% of women older than 35 years[1]. The unusual locations are rare and may be seen including primary retroperitoneal growth; its incidence is estimated at 1.2%[2]. This rare entity challenges in pre-operative diagnosis even with diagnosis imaging. We present a case of large posterior retroperitoneal cellular leiomyoma in a postmenopausal woman.

Case Report

A 65-year-old female presented with vaginal mass incidentally found by physician from a routine check up one year ago. She denied further investigation and treatment. On Aug 2014, she complained of minimal vaginal bleeding and came to the outpatient gynecological clinic of our hospital. There was no urinary or bowel symptoms, pelvic pain, nausea, vomiting, anorexia and weight loss. She had one child with two spontaneous first trimester miscarriages. Her menopausal age was 55 years old without a history of hormonal replacement therapy taking. There was no significant past medical history except ampicillin allergy. She underwent one cesarean section 27 years ago. Her general condition was stable, no pallor and no palpable abdominal mass. The vaginal examination revealed a posterior mass protruding from right posterolateral wall of vagina. Its size was more than 6 cm in diameters. The upper border of mass could not be identified. The mass had smooth surface, not tender and rubbery in consistency. The vaginal mucosa was normal for this age. The cervix was difficult to palpable because of the constrictive effect of the mass. The uterus and right adnexae was hardly to identify. However, the left adnexae was normal by bimanual examination. Rectal examination revealed a bulging anterolateral mass on the right side with normal and smooth rectal mucosa.

The computed tomography contrast was performed to identify the origin, the nature of mass and the possibility of resection. The result showed a well-defined lobulated heterogeneous enhancing mass with internal calcification in pelvic cavity size 6.5x7.5x7.9 cm in AP, transverse, and longitudinal dimension, respectively. The mass abutted lower segment of uterus cervix and vagina. It had pressure effect to uterus and rectum. The fat plain differentiation between mass and

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the rectum was seen (Fig. 1). The density and location of the mass from CT suggested subserosal cervical leiomyoma. However, a malignant condition could not be excluded. CA125, CA19-9, AFP, Beta hCG and LDH were in normal values. The laparotomy was performed via low midline incision. Right posterior retroperitoneal mass was identified at the level of internal cervical os to the upper part of vagina. The mass was round shape, gray white color, rubbery to firm consistency, 8 cm in diameters without connection to uterus and cervix. It compressed vaginal lumen posteriorly but not involved into vaginal and rectal canal. The outer surface is nodular and partially covered by fibrous tissue. Cut surface showed lobulated light yellow non-homogeneous tissue with necrosis and calcification (Fig. 2). The uterus, bilateral adnexa, rectum and urinary bladder appeared normal. The operation was total hysterectomy, bilateral salpingoophorectomy and tumor mass resection. The mass was difficulty dissected from the surrounding tissue but was entirely resected without injury to adjacent organs. The hemostasis was achieved.

Microscopic description of mass showed cellular spindle cell tumor with well-defined border. The tumor cells showed no definite atypia and no increasing of mitotic activity (less than 3 per 10 HPF) (Fig. 3). Some areas of tumor necrosis and hyalinization were seen. No invasive lesion was present. All resected margins were free as shown in Fig. 3. Pathological diagnosis revealed spindle cell tumor. The diagnosis was cellular leiomyoma. The uterus showed atrophic endometrium with unremarkable myometrium and both salpinx. Incidental finding of endocervical polyp was found.

The patient recovered and discharged from hospital on the fourth postoperative day. She returned to follow-up 1, 4 and 12 weeks after the operation without complications and recurrence.

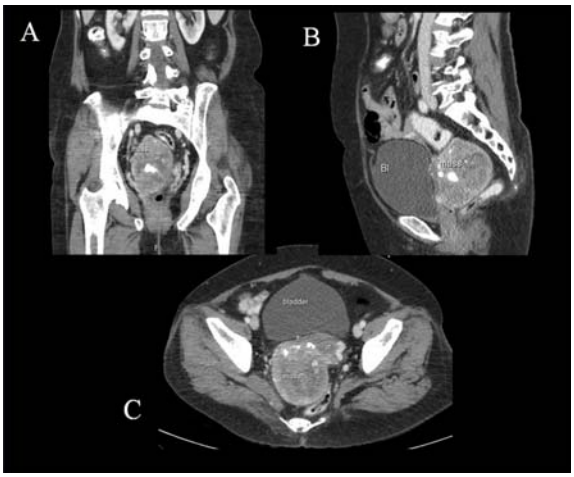

Fig. 1 Coronal view (A), sagittal view (B) and transverse view (C) of CT contrast imaging.

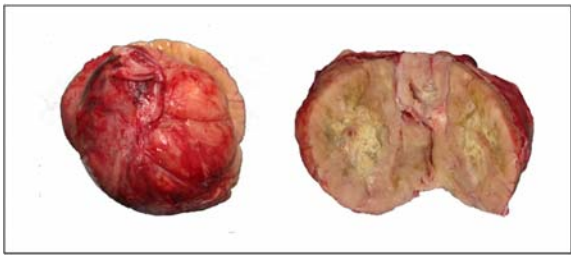

Fig. 2 Retroperitoneal mass after resection.

Discussion

Primary retroperitoneal cellular leiomyomas are rare and only seen in case reports. Poliquin et al reported 105 cases of retroperitoneal leiomyoma's features. The mean age was 46.27 years. One third of the cases had a previous hysterectomy, myomectomy or had concurrent uterine leiomyoma during investigation[2]. Pre-operative diagnosis was difficult because of the rarity and the non-specific manifestation. It may be asymptomatic and detected incidentally at a routine check up or cause abdominal fullness, urinary retention, constipation, fatigue, weight loss and pelvic pain[3-6]. Additionally, unusual manifestation included a perineal hernia swelling in the perineal region of recent onset and sexual intercourse difficulty had been reported[7,8].

Pre-operative imaging of ultrasonography (USG), computed tomography (CT) or magnetic resonance imaging (MRI) are necessary to assess the tumor location and its relation to other pelvic organs. USG may depict a well-defined homogeneous echo-texture mass within retroperitoneal position. The typically imaging of leiomyoma seen in CT is homogeneous attenuation mass. MRI is the most reliable technique for evaluating retroperitoneal mass and may also help in differentiating between sarcomatous and leiomyoma[9,10]. In this case, gadolinium-enhanced diethylenetriaminepenta-acetic acid (Gs-DTPA) dynamic MRI was not performed before operation.

Retroperitoneal smooth muscle tumors are usually malignant. It is not possible to rule out malignancy based on imaging features alone. Tumor markers are neither sensitive nor specific for

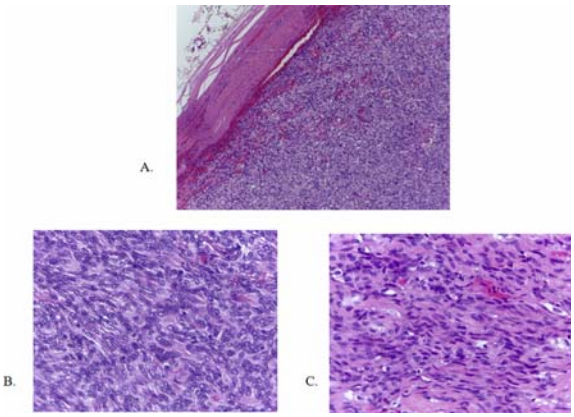

Fig. 3 Microscopic findings.At low power (A), the tumor is sharply demarcated to the surrounding tissue and shows a hypercellular proliferation of spindle cells, arranging in a loose, haphazard, and storiform pattern (H&E stain, X4). At high power (B, C), the tumor is composed of spindle cells with bland cytology. They have variable amounts of cytoplasm and fusiform nuclei. The hypercellular areas are separated by bands of collagenized stroma. Nuclear atypia and mitotic figure are not present (H&E stain, X40).

sarcomatous process[2]. Therefore, complete resection with pathological examination is mainstay of treatment. Radiologically guided biopsy may be helpful and use for asymptomatic patients refusing operation, but depending on the location of the tumor.

The most common location is in posterior pelvic peritoneum including rectovaginal septum, pararectal space and the pouch of Daouglas. Histologically, most have low mitotic activity (<10/10 HPF) and rare nuclear atypia. One-third of cases present with hyaline degeneration. The prognosis seems to be favorable. From systematic literature review of Poliquin, 5 of 105 cases had recurrent after an average period 30.5 months. Two cases of recurrence described positive margins at the time of initial surgery. Another three cases were subsequently reclassified as leiomyosarcoma. Survival of leiomyosarcoma cases were achieved only in cases who underwent radiation therapy[2].

There was no previous study mentioned about the cellularity of retroperitoneal leiomyoma. Cellular leiomyomas are significantly more cellular than the normal myometrium, but lack nuclear atypia, tumor cell necrosis or increased mitotic activity (mitotic rate is usually less than 4/10 HPF). The cells are small and round to spindle shaped. The natural history is the same as typical leiomyomas. Cytogenetically, cellular leiomyoma has been found to be accompanied by loss of almost the entire short arm of chromosome 1. The cellular leiomayoma must be distinguished from leiomyosarcoma (malignant) and endometrial stromal neoplasm (benign).

The etiology and pathogenesis of retroperitoneal leiomyoma remain unclear. There are many theories regarding the origin of retroperitoneal tumor. Zaitoon suggested parasitic fibroid theory. The pedunculated subserosal leiomyomas lost their blood supply and grew into retroperitoneum[11]. Kho and Nezhat reviewed 12 cases of retroperitoneal leiomyomas and proposed an iatrogenically created after surgery as the cause, particularly surgery using morcellation techniques[12]. Stutterecker described two cases of isolated leiomyomas in pubovesicle space and claimed that embryonal remnants or local vessel musculature might be possible origins with hormonal influences as cofactors for development of their tumors[13]. Another theory described by Watanabe was mesometrial smooth muscle growth beneath the peritoneal membrane. He founded that retroperitoneal myoma and mesometrial smooth muscle had been similar both anatomic and immuno-histochemical characteristics[14].

This case showed an isolated mass abutted the cervix. Pathological examination revealed calcified leiomyomata that often found in pedunculated subserosal tumor[1]. The concept of parasitic origin was the most accepted explanation in this case.

Conclusion

Retroperitoneal leiomyoma is a rare neoplasm with limited evidence regarding etiology, nature of tumor, recurrence rate and uncertain malignant potential. It is difficult to establish the exact nature of a retroperitoneal mass preoperatively. Imaging may be helpful to determine the nature, location, and also distinguishing malignant conditions. Unfortunately, there was no accurate imaging to exclude sacromatous change. Complete surgical excision with pathological diagnosis is required.

What is already known on this topic ?

Retroperitoneal myoma is rare neoplasms. It can be asymptomatic or can cause pelvic discomfort, urinary frequency, abdominal fullness or back pain. It presents diagnostic and therapeutic challenges. Final diagnosis is made intra-operatively on direct visualization. Complete excision is the mainstay of treatment. The etiology and pathogenesis are still

obscure.

What this study adds ?

At this time, there have been no reports of retroperitoneal cellular leiomyoma case in Thailand. (This search using the keywords 'extrauterine leiomyoma' 'retroperitoneal leiomyoma' and 'Thailand' from Pubmed, SCOPUS, OVID medline and Thai Index Medicus). This case presented with posterior vaginal mass which difficult to establish the exact nature of tumor pre-operatively. From the result of CT suggested subserosal cervical leiomyoma that also miss the exact site of the mass. This case showed an isolated mass abutted the cervix and pathological examination revealed calcified leiomyomata that often found in pedunculated subserosal tumor. The concept of parasitic origin is the most accepted explanation in our case report.

Consent

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

Acknowledgement

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Potential conflicts of interest

None.

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รายงานผู้ป่วย: เนื้องอกมดลูกด้านหลังเยื่อบุช่องท้องในอุ้งเชิงกราน

ฐานิตรา ตันติเตมิท, สุทธา หามนตรี, ลิขิต รังสิรัตนกุล, เมสิตา สุขสมานวงวศ์

เนื้องอกมดลูกเป็นโรคเนื้องอกชนิดไม่ร้ายแรงทางนรีเวชที่พบได้บ่อย โดยเฉพาะบริเวณกล้ามเนื้อมดลูก ในตำแหน่งอื่นอาจเกิดเนื้องอกมดลูกได้แต่พบได้น้อย เช่น บริเวณหลัง เยื่อบุช่องท้อง ส่งผลให้วินิจฉัยและรักษาได้ยากรายงานนี้นำเสนอผู้ป่วยหญิงอายุ 65 ปี มาพบแพทย์ด้วยก้อนบริเวณด้านหลังเยื่อบุช่องคลอด ตรวจเอกซเรย์คอมพิวเตอร์พบก้อนในอุ้งเชิงกรานขนาด 6 เซนติเมตร ติดกับบริเวณมดลูกด้านล่าง ปากมดลูกและช่องคลอด ให้การวินิจฉัยเบื้องต้นว่าเป็นเนื้องอกที่บริเวณกล้ามเนื้อปากมดลูกชั้นนอกสุด ในระหว่างการผ่าตัดพบว่าก้อนที่ตรวจพบดังกล่าวแยกจากมดลูกชัดเจน อยู่ในระดับเดียวกับปากมดลูก บริเวณด้านหลังเยื่อบุช่องท้อง ผลการตรวจทางพยาธิวิทยายืนยันว่าเป็นเนื้องอกมดลูก

ผู้ป่วยได้รับการผ่าตัด มดลูก รังไข่ และก้อนเนื้องอกออกทั้งหมดโดยไม่มีภาวะแทรกซ้อนใดๆ เนื้องอกมดลูกที่บริเวณหลังเยื่อบุช่องท้องเป็นภาวะที่พบได้น้อย ข้อมูลจากประวัติ การตรวจร่างกาย การตรวจภายในรวมถึงการส่งตรวจทางรังสีต่างๆ ไม่สามารถให้การวินิจฉัยเบื้องต้นได้ชัดเจน การผ่าตัดเพื่อเอาก้อนออกเพื่อส่งตรวจทางพยาธิวิทยาจึงเป็นวิธีที่จำเป็นในการวินิจฉัยและรักษาภาวะดังกล่าว