Case Report

Fallopian tube lipoleiomyoma with degeneration: a case report and literature review

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Abstract: Lipoleiomyoma is a rare, benign leiomyoma variant. It is relatively common in the uterine area of the female reproductive system but rare in other areas and has not been reported in the fallopian tubes. In this paper, we report a perimenopausal woman with a lipoleiomyoma arising from the ampulla of the fallopian tube with hydropic degeneration. What makes this case even rarer is the combination of hydrops of the pelvis and the abdominal cavity. Microscopically, the tumor was composed of smooth muscle tissue mixed with varying amounts of mature adipose tissue. The immunohistochemical markers were Des(+), SMA(+), H-caldesmon(+), ER(+), CD34(-), HMB45(-), Melan-A(-), CD10(-), S-100 focal adipocyte (+), and the positive rate of Ki67 was about 1%. Through this case report and review of similar literature, we hope to improve the understanding of the diagnosis and treatment of fallopian tube smooth muscle-derived tumors.

Keywords: Fallopian tubes, lipoleiomyoma, hydropic degeneration, hydrops

Introduction

The fallopian tubes are an important part of the female internal reproductive organs and play an important role in the conception process. The internal reproductive organs all originate from the mesonephric ducts or the Mullerian ducts. Therefore, all the tumors that can occur in the uterus and ovaries can also theoretically occur in the fallopian tubes, so there are many kinds. However, the incidence rate of fallopian tube tumors is very low, and benign tumors are even rarer. Among them, adenomatoid tumors are relatively common [1], but others such as leiomyoma are extremely rare [2]. Lipoleiomyoma, also known as lipomatous leiomyoma, is a special histological subtype of leiomyoma and is composed of benign smooth muscle components and mature adipose components. It mainly occurs in the uterus of the female reproductive system [3, 4], but it is occasionally reported in the ovaries, the broad ligament, the vulva, and other areas [5-7]. This is the first case of fallopian tube lipoleiomyoma with hydropic degeneration and a small amount of hydrops of the pelvis and abdominal cavity.

Clinical data

A 48-year-old premenopausal woman was hospitalized 15 days previously for acute pain in her right lower abdomen. The examinations done on admission were as follows: (1) Ultrasound: localized adenomyosis with multiple leiomyomas; hyperechoic inhomogeneous masses about 12.3 × 11.7 × 6.5 cm in size were found in the right lower abdominal cavity. The boundary was clear and the shape was elliptic, and there was no significant adhesion with the uterus, the bilateral ovary, or the intestinal canal. There was a small amount of hydrops of the pelvis and abdominal cavity, suggesting that it is possible to consider an ovarian origin (Figure 1A, 1B). (2) Abdominal enhancement CT examination: multiple myomas of the uterus, irregular soft tissue masses of 11.1 × 9.1 cm with clear boundaries were found in the right abdominal and pelvic cavities, a small amount of hydrops of the abdominal and pelvic cavities, suggesting that an ovarian origin should not be considered, and a further examination was recommended to determine the specific sources (Figure 1C, 1D). (3) Specialty
examination: a solid mass about 10 cm in size was found in the right lower abdomen palpates, which was active and closely related to the uterus. We also observed anteversion of the uterus with an irregular shape, the size of a woman four months pregnant. (4) Tumor mark-
ers: NSE 21.32 ng/ml, normal. The patient had normal menstruation, with no irregular vaginal bleeding, abnormal drainage, or other changes. She had a history of uterine leiomyoma exfoliation from more than 20 years ago, with no other medical history or any family genetic history.

Tentative clinical diagnosis: the nature of the pelvic mass to be investigated, considering an ovarian malignant tumor? Then a laparoscopic exploration and an open right fallopian tube tumor resection were performed. Intraoperative findings: solid tumor of the right fallopian tube, about 12 × 9 × 8 cm in size, enveloped outside. The fallopian tube was elongated and separated from the right pelvic cavity with no adhesion to the surrounding tissues (Figure 2A). A frozen section examination was performed during the operation. The results suggested a
benign spindle cell tumor. The patient's condition was explained to the family who did not want the extent of surgery to be expanded.

Materials and methods

The specimens from the fallopian tube masses were frozen, sectioned, and stained rapidly during the operation. The conventional specimens were fixed with 4% neutral formaldehyde, the materials were selected conventionally, paraffin embedded, sectioned, and HE stained. The EnVision two-step method was adopted for the immunohistochemical labeling. Antibodies used: Des, SMA, H-caldesmon, ER, S-100, CD34, HMB45, Melan-A, CD10, and Ki67. The antibodies were purchased from Roche.

Pathological analysis

General description: An elliptic mass of 14 × 9 × 7 cm in size with a complete capsule. Most of the sections were gray-white, and some of the areas were gray-white and gray-yellow, tough and faintly nodular. A fallopian tube was attached to one side of the mass, 7.5 cm long and about 0.3-0.5 cm in diameter. The fimbria and ampulla of the fallopian tube were closely adhered to the masses, compressed and flat in shape, and the isthmus was relatively free (Figure 2B).

Microscopic appearance: The tumor was most closely related to the tubal wall of the ampulla of the fallopian tube, and there was no significant hypertrophy in the lamina propria of the fallopian tube. The tumor was mainly composed of smooth muscle cells with a mild morphology and various amounts of mixed mature adipocytes distributed unevenly, with hydropic degeneration in the partial interstitium. Immunohistochemistry: Des(+), SMA(+), H-caldesmon(+), ER(+), CD34(-), HMB45(-), Melan-A(-), CD10(-), S-100 focal adipocyte (+), and the Ki67 positive rate was about 1% (Figure 3A-F).

Final pathological diagnosis: Right fallopian tube lipoleiomyoma with hydropic degeneration.

Discussion

A primary tumor of the fallopian tube originating from the fallopian tube or the mesosalpinx is a rare gynecological tumor, and a benign tumor is even rarer. According to the origin of the Mullerian cells, the tumors are mainly classified as epithelial tumors, mesodermal tumors, endothelioma, or teratoblastoma. Fallopian tube leiomyoma is a rare type of mesodermal tumor, and lipoleiomyoma is an even rarer type of leiomyoma, and its clinical manifestations are similar to those of ordinary...
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leiomyoma of the fallopian tubes. CT images of such tumors are often misdiagnosed as ovarian in origin, and the ultrasound images are of relatively great significance, for they can show the separation relationship between the tumor and the ipsilateral ovary [8], and MRI has a better diagnostic value when necessary [9]. However, it is difficult to identify the origin of fallopian tube tumors through imaging examinations, and one cannot distinguish whether the tumors are benign or malignant. The preoperative detection of tumor markers has a certain suggestive effect, but ultimately it depends on pathology to determine the nature of the tumor.

The patient in this case was a perimenopausal middle-aged woman with no abnormal menstruation who visited the doctor for acute pain in her right lower abdomen. Through the relevant examinations, a solid tumor of more than 10 cm was found in the right abdomen and pelvis, but it was difficult to determine the specific ovarian or fallopian tube source using CT. Because the incidence of ovarian tumors is significantly higher than fallopian tube tumors, and because this case was complicated by a small amount of hydrops of the pelvis and abdominal cavity, the clinical preoperative suspicion was ovarian cancer, even though the tumor markers were in the normal range, except for NSE. During the clinical operation, it was found that a tumor was located in the fallopian tube and it was rapidly frozen and sectioned during the operation. Confirming the benign nature of the tumor was crucial to guiding the direction of the clinical operation. The sections and histopathological morphology of the lipoleiomyoma of the fallopian tube were similar to those of other invasion sites. The gross specimen of the tumor showed a close relationship with the fallopian tube, and there was an outer capsule. Under the microscope, the tumor presented a mixed morphology of typical lipoleiomyoma and adipose tissue with some interst-
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Tial degeneration. Histology suggested that the tumor originated from the ampulla of fallopian tube with no significant hyperplasia in the myosalpinx. Combined with the intraoperative observation and the pathology, it was confirmed to be a right fallopian tube lipoleiomyoma with hydropic degeneration. The special feature of this case is the combination of a small amount of pelvic and abdominal hydrops, which may be an important factor leading to a clinical misdiagnosis. The postoperative follow-up showed that the prognosis of the patient was good, and the effusion disappeared (Figure 4A, 4B). It is speculated that the formation of the hydrops may be related to the tumor, but the mechanism is not yet clear, and this inference needs to be further supported by the long-term follow-up data.

As far as we know, this is the first case of lipoleiomyoma in the fallopian tube. Because the embryological origin of the fallopian tube is the same as that of the uterus and other parts, referring to the related literature of leiomyomas in the fallopian tube and lipoleiomyoma in the uterus and other parts, it is speculated that the smooth muscle cells in the lipoleiomyoma of the fallopian tubes may originate from the myosalpinx or the vascular cells supplying the fallopian tubes [10]. Adipocytes may originate from mesenchymal cells with a multidirectional differentiation potential or adipocytes transformed by smooth muscle cells [4]. Compared with the uterus and other sites, fallopian tube site smooth muscle-derived tumors are extremely rare, and it has been suggested in the literature that this may be related to the insensitivity of the fallopian tube smooth muscle to estrogen [8]. In 1976, Hone et al. proposed that the pathogenesis of leiomyoma is different from that of uterine tumors and may be similar to gastrointestinal leiomyoma, based on the histomorphological characteristics of no hyperplasia and hypertrophy of the intrinsic myometrium of the fallopian tubes in leiomyoma [11]. But in 2018, Li et al. found that there was a high ratio mutation of exon 2 of MED12 in leiomyoma of adnexa and the uterus using gene detection, which revealed that the pathogeneses of oviduct and uterine leiomyoma were similar, further supporting the viewpoint of different gene changes in pelvic and extra-pelvic smooth muscle tumors [12]. At present, the pathogenesis of this tumor is still not clear, so further research is needed. This kind of tumor can occur in any part of the fallopian tube, often solitary, mainly in the isthmus, and rarely in the ampulla. Because the fallopian tube is relatively free and the blood supply is not rich, when the tumor is large, it can appear with changes such as torsion or degeneration [13, 14]. However, most of the tumors are small, without

![Figure 4. A, B. Ultrasound images at 3 months and 11 months after the operation: The right ovary can be seen, and part of the right attachment area is shown, and there is no significant abnormal echo or free fluid dark area.](image-url)
special symptoms or signs, and they are often ignored or only occasionally found. Some patients seek medical treatment because of large tumors or acute pain in the abdomen. It is difficult to determine the origin of fallopian tube tumors using general imaging examinations, which makes preoperative diagnoses difficult. Intraoperative explorations and pathological examinations are the basis of the diagnosis. Surgical resection is the most effective treatment, and the prognosis is good. To sum up, this case enriches the clinical diagnosis understanding of female pelvic and abdominal masses and broadens the diagnostic field of the fallopian tube smooth muscle-derived tumors, so it has a diagnostic significance.

Disclosure of conflict of interest

None.

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