

## CASE REPORT

# Ovarian leiomyoma accompanied by a neglected abdominal wall leiomyoma: A case report and literature review

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**Key Clinical Message**

Ovarian leiomyoma and abdominal wall leiomyoma are both rare clinical entities. Here, we report the rare case with ovarian leiomyoma accompanied by a neglected abdominal wall leiomyoma to raise the awareness of clinicians of ovarian leiomyoma and multiple occurrences of benign leiomyoma for appropriate diagnosis.

**KEYWORDS**

abdominal wall leiomyoma, diagnosis, ovarian leiomyoma, pathogenesis

## 1 | INTRODUCTION

Ovarian leiomyoma has a rare incidence, which is thought to account for 0.5%–1% of all benign ovarian tumors.<sup>1,2</sup> So far, fewer than 200 cases have been reported since it was first described in 1862.<sup>3</sup> There are no specific clinical manifestations, tumor markers, or imaging features of ovarian leiomyoma. Preoperative diagnosis is really difficult and postoperative histopathological examination is the golden diagnosis. There is still no consensus on the origin of ovarian leiomyoma. Some articles present that ovarian leiomyoma always accompanied by a leiomyoma of the uterus.<sup>4</sup> The pathogenesis of abdominal wall leiomyoma remains unclear. Some theories declare that it may originate from the uterus.<sup>5</sup> Others claim that it is benign metastasizing leiomyoma resulting from myomectomy or hysterectomy for uterine leiomyoma.<sup>6</sup>

## 2 | CASE PRESENTATION

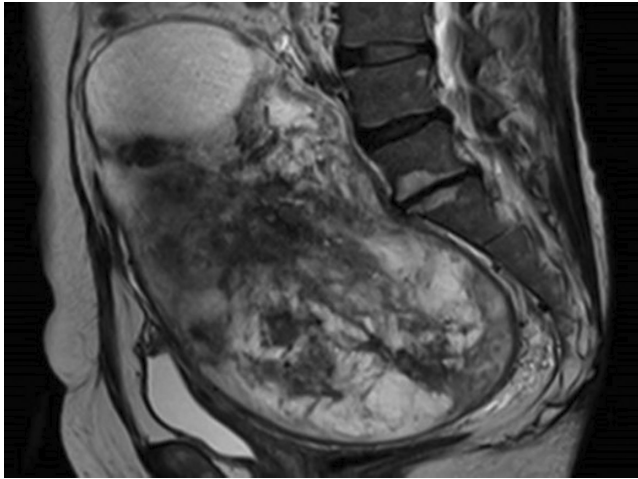
A 49-year-old woman presented at our department with a complaint of a huge abdominal mass. One year before her

presentation to our hospital, the mass was about fist-sized, as she said. On clinical examination, there was a huge hard mass occupying the pelvic cavity, with the upper boundary reached the umbilical three fingers, both sides reached the axillary front, clear boundary, poor motion, and no tenderness. On further investigation, magnetic resonance imaging (MRI) revealed a large irregular soft tissue mass with cystic degeneration and dense adhesion (Figure 1). The left ureter is compressed by the tumor and the proximal part of it is swollen. Serum carbohydrate antigen-125 and carbohydrate antigen-199 levels were elevated at 128 U/mL (0–35 U/mL) and 48 U/mL (0–35 U/mL), respectively.

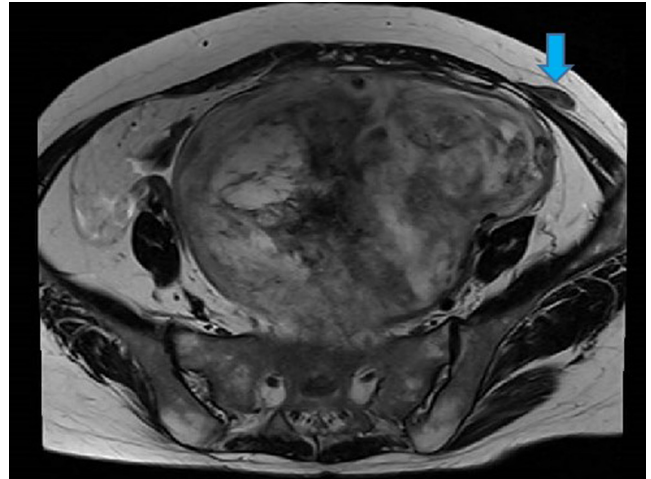
Ovarian cancer cannot be excluded before surgery. Exploratory laparotomy was performed, during which a solid tumor with cystic component on the left ovary was identified. The ovary was enlarged, hard, and full in shape. The patient underwent laparotomy left salpingo-ovariectomy. Postoperative pathology with hematoxylin and eosin (H&E) stain showed spindle cell (Figure 2), confirmed by histopathological examination: SMA (+) (Figure 2), desmin (+) (Figure 2), CK(–), Ki67(+), CD34(–), and inhibin(–).

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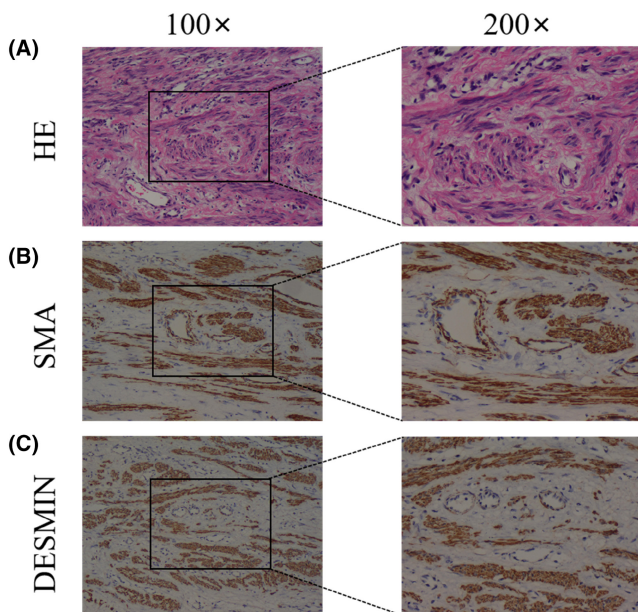
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**FIGURE 1** MRI illustrates a huge pelvic mass with mixed signals on T2WI image.



**FIGURE 3** MRI shows the lump in the left abdominal wall with low signal, arrow marks the location.



**FIGURE 2** Histopathological images of the ovarian leiomyoma. (A) HE stain shows spindle cell; (B) SMA; (C) desmin.

Fifty days later, she touched a thumb-sized lump in the left abdominal wall. Retrospectively, we find out the pelvic MRI imaging taken before the previous surgery. A small regular mass did exist in the left abdominal wall accompanied with the ovarian leiomyoma (Figure 3). It was about the size of the thumb abdomen, with clear boundaries, good motion, and without tenderness.

Recognizing the multiple occurrences of benign leiomyoma, we told her that leiomyoma may occur in other organs someday. We told the patient the abdominal mass could be observed temporarily. But she feared that the abdominal mass was malignant. She occasionally felt pain. She could not bear the huge psychological pressure and determined to



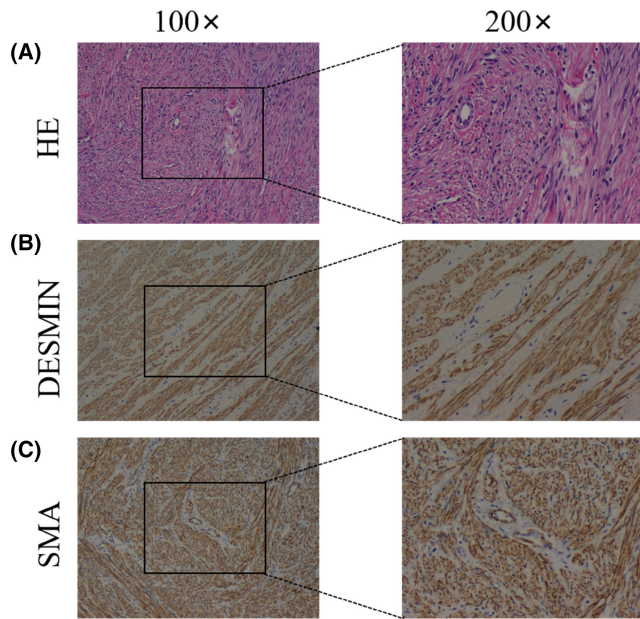
**FIGURE 4** Postoperative specimen of the abdominal wall leiomyoma.

have another operation for the postoperative pathology. The abdominal wall lump was cutoff outside the aponeurosis of obliquus externus abdominis (Figure 4). Pathology with H&E stain also showed spindle cell (Figure 5), confirmed by histopathological examination: Desmin (+) (Figure 5), SMA(+) (Figure 5), CK(−), vimentin (−), Ki67 (5%+), CD34 (blood vessel+), and S-100 (−).

Follow-up: There was no evidence of recurrence or emerging leiomyoma during 2 years of follow-up.

### 3 | DISCUSSION

This report present a case of ovarian leiomyoma accompanied by a neglected abdominal wall leiomyoma. The two leiomyoma could have been cured with one operation. Clinicians need to improve the recognition



**FIGURE 5** Histopathological images of the abdominal wall leiomyoma. (A) HE stain shows spindle cell; (B) Desmin; (C) SMA.

of ovarian leiomyoma and be alert to the presence of leiomyoma in other organs. There is still no consensus on the origin of ovarian leiomyoma. Some scholars classify that it may originate from the blood vessels in the hilum of the ovary and the smooth muscle of the ovarian ligament,<sup>7</sup> whereas others point to the ovary cortex, ovary stroma<sup>8</sup> and other smooth muscle components in ovarian tumors. It is reported that about 78% of ovarian leiomyoma were associated with leiomyoma of uterus and they may have a common trigger.<sup>9</sup>

The pathogenesis of abdominal wall leiomyoma remains unclear. The patient in our case underwent laparoscopic total hysterectomy for uterine leiomyoma 6 years ago without using myoma crusher. The abdominal wall leiomyoma was not present at the original laparoscopic puncture site. So we highly suspect that the abdominal wall leiomyoma was the result of hematogenous metastasis, rather than implantation metastasis. This view is consistent with Yuan<sup>6</sup> who claimed that benign metastasizing leiomyoma occurs in women with a history of myomectomy or hysterectomy for uterine leiomyoma. Meanwhile, Brincat<sup>10</sup> reported a benign metastasizing leiomyoma of the lung with a history of uterine leiomyoma without surgery, suggesting a multifocal histogenesis rather than metastasis. Two Chinese patients have been reported for the abdominal wall leiomyoma originated from vessel containing smooth muscle in the abdominal wall and mesenchymal tissue of peritoneum. The primary abdominal leiomyoma was not excluded in this case.

Ovarian leiomyoma exhibit no specific clinical symptoms. So it is seldom diagnosed before surgery. The majority of ovarian leiomyoma are unilateral small and

regular shaped. There is a rare report of huge ovarian leiomyoma up to 30 cm.<sup>11</sup> Patients with large tumor may experience the corresponding symptoms due to the compression of the huge pelvic mass, others may encounter Meigs syndrome with hydrothorax and ascites.<sup>12</sup> The huge leiomyoma in this case gave a big pressure on adjacent organs, resulting in ureterectasis and dense adhesion. Ultrasound and MRI cannot distinguish them from other ovarian tumors. Ultrasound usually report regular solid mass with clear boundary like any other benign tumors. Some may show the typical swirling structure of leiomyoma. We should distinguish ovarian leiomyoma from ovarian fibroma, ovarian sex cord stromal tumor, and leiomyosarcoma. Although MRI has advantages over computed tomography (CT) in soft tissue resolution, the MRI findings of ovarian leiomyoma and ovarian sex cord stromal tumor lack specificity.<sup>13</sup> Studies have shown that the MRI signals of primary ovarian leiomyoma are similar to those of uterine leiomyoma.<sup>14</sup> There have been no sensitive serum tumor markers for differential diagnosis. Intraoperatively we should distinguish ovarian leiomyoma from intravascular leiomyoma, ovarian involvement in peritoneal disseminated leiomyoma.

The diagnosis can only be confirmed by pathological tests and depends on immunohistochemistry. Smooth muscle actin (SMA) and Desmin are both mesenchymal tissue derived markers. SMA is used in the diagnosis of smooth muscle tumors. It is used to identify leiomyoma and exclude follicular membrane cell tumor. Desmin can maintain the migration direction of actin and myosin filaments, expressing in almost all smooth muscle cells. A positive desmin can exclude fibroma. Vimentin is a kind of intermediate fibrous protein in eukaryotic cells and highly expressed in mesodermal cells. It is positive in both leiomyoma and fibroblast. Inhibin is a kind of glycoprotein hormone, produced by the preovulation follicle and the corpus luteum. Almost all ovarian granulosa secretes inhibin.  $\alpha$ ,  $\beta$ A, and  $\beta$ B are subunits of inhibin, produced by granulosa cell tumor of ovary, being negative in leiomyoma. At the same time, characteristics such as activity of mitosis, cytological heteromorphism, and tumor necrosis should be considered to distinguish leiomyoma and leiomyosarcoma.<sup>15</sup> In this case, the immunohistochemistry of SMA (+), desmin (+), and inhibin (–) confirmed that the tumor was derived from smooth muscle tissue rather than sex cord stromal tumor such as granulosa cell tumor or fibroma.

In terms of treatment, ovarian leiomyoma is mainly treated by surgery, with the comprehensive consideration of the patient's age, fertility need, tumor size, contralateral ovary and other factors. Resection of the affected adnexa is often performed, which is suitable for those who have no fertility requirements or large tumor with heavy adhesion. For young patients with fertility requirements,



ovary-preserving surgery can be performed. There is usually no recurrence, even in active or atypical leiomyoma. Our patient in this case was a perimenopausal woman with huge ovarian leiomyoma, which occupied almost the whole ovary. The affected side adnexectomy was performed for radical treatment.

## 4 | CONCLUSION

We should consider ovarian leiomyoma as one of the differential diagnosis when patients present with solid ovarian mass, especially accompanied with uterine leiomyoma or having a history of uterine leiomyoma. At the same time, we should be alert for leiomyoma in other organs. Ovarian leiomyoma exhibit no specific symptoms. It is seldom diagnosed before surgery. The diagnosis can only be confirmed by pathological tests and immunohistochemistry.

## AUTHOR CONTRIBUTIONS

**Jing Li:** Validation; writing – original draft; writing – review and editing. **Mingxing Sui:** Data curation; writing – review and editing. **Xiaodan Sun:** Investigation; writing – review and editing.

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## CONFLICT OF INTEREST STATEMENT

The authors declare that there is no conflict of interests for this article.

## CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

## DATA AVAILABILITY STATEMENT

All data generated or analyzed during this study are included in this article. Further enquiry can be directed to the corresponding author.

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