

Case Report

Mature cystic teratoma of the uterosacral ligament successfully treated with laparoendoscopic single-site surgery

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Abstract

Objective: Although the majority of teratomas are encountered in the ovary, extragonadal mature cystic teratoma is an unusual disease entity, and the most common site is the omentum.

Case Report: The occurrence of this tumor on a uterosacral ligament is extremely rare with enigmatic etiology. To our knowledge, there have been only three cases reported to date that describe a mature cystic teratoma of the uterosacral ligament, and this is the first report of successful treatment of these rare tumors with laparoendoscopic single-site surgery (LESS).

Conclusion: In the present study, we report a mature cystic teratoma of the uterosacral ligament successfully treated with LESS in a 34-year-old woman with a preoperative diagnosis of mature cystic teratoma of the left ovary.

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Keywords: laparoendoscopic single-site surgery; mature cystic teratoma; uterosacral ligament

Introduction

Teratomas are neoplasms composed of tissues from all germinal layers with unknown histogenetic origin. Although these tumors most commonly occur in the ovary, a number of other locations were documented, equivalent in frequency to 0.4% of all ovarian teratomas [1]. These extragonadal teratomas occur most commonly in the omentum [2] and less commonly in the fallopian tube, uterus, diaphragm, liver, mediastinum, and thymus. Specifically, the teratomas of the uterosacral ligament are exceedingly rare, and only three cases have been described to date [1,3,4]. Accordingly, our report may be helpful to evaluate the clinical nature and surgical management of this rare tumor.

This is the first report of the successful treatment of a mature cystic teratoma of the uterosacral ligament with laparoendoscopic single-site surgery (LESS).

Case report

A 34-year-old, gravida 3, para 2 woman with no disease history was referred to our outpatient clinic for an asymptomatic pelvic mass detected on routine checkup. On the physical examination, a fixed soft mass was palpated between the left adnexa and cul-de-sac. The laboratory data including tumor markers such as CA 19-9 and CA 125 were within normal limits. Ultrasonography and computed tomography (CT) indicated a normal-sized right ovary. However, a round mass measuring 4.0 × 3.2 cm was seen in the left posterior pelvic cavity (Fig. 1). The cyst was observed to contain some internal debris, fat-density material, and distinct calcification, which were findings consistent with those of a mature cystic teratoma.

With the diagnosis of a mature cystic teratoma, possibly originating from the left ovary, LESS was performed with

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Fig. 1. Computed tomography (CT) scan of the tumor (arrow), which shows internal debris and fat component.

a 15 mm intraumbilical incision using a device described in our previous report [5]. Under laparoscopic observation, a soft white mass was found attached to the left uterosacral ligament (Fig. 2). The left ovary with a streaky appearance was observed in the normal anatomical position distant from the mass, and the right ovary was revealed to be of normal size and shape (Fig. 3). The mass had no feeding vessel or ligamentous connection to other organs except for the attachment of fine vessels to the left uterosacral ligament. The overall procedures performed in this case were similar to those performed in conventional laparoscopic surgery that uses conventional laparoscopic instruments. After the careful excision of the mass from the uterosacral ligament using curved dissector forceps and electrosurgical scissors, it was removed through the umbilical incision using an EndoPouch (Ethicon Endo-Surgery, Cincinnati, OH, USA). Once the mass was extracted, the bed was carefully inspected and bleeding areas were secured using bipolar electrocoagulator. The umbilical incision was repaired postoperatively and hidden completely, and the patient recovered uneventfully and was discharged 2 days after the surgery.

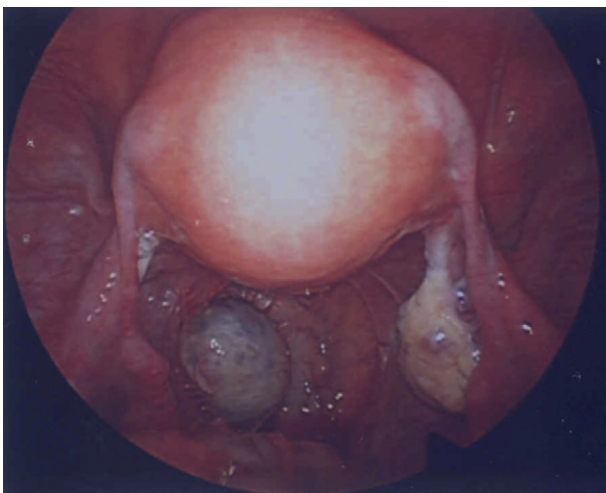


Fig. 2. Laparoscopic view of the pelvis. The mass is located on the left uterosacral ligament and is not connected to other organs.

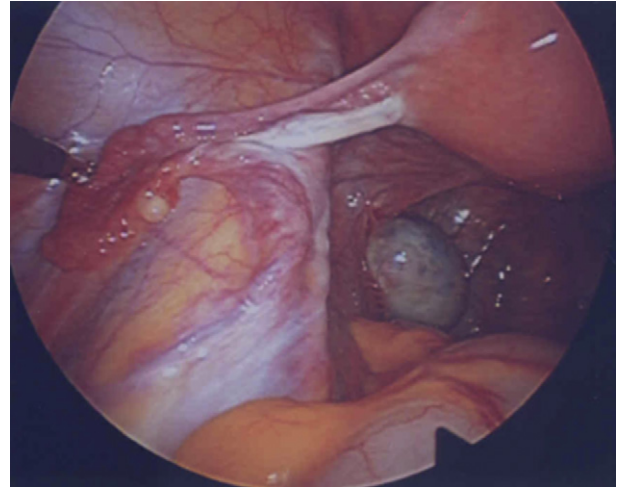


Fig. 3. Laparoscopic view of the pelvis. Note that the left ovary was streaky and located in the normal anatomical position distant from the mass.

Macroscopic histology of the mass revealed a cystic mass measuring 5×4 cm, filled with sebaceous material, matted hair, and teeth, confirming the pathological diagnosis of a benign mature cystic teratoma. Ovarian tissues such as ovarian stroma and corpus albicans were not found, and the cyst wall was lined by skin tissue including epithelium and collagen fibers.

Discussion

Although teratomas arise mainly from totipotential gonadal cells, extragonadal teratomas have been reported occasionally, mostly along the midline [1]. However, teratomas of the uterosacral ligament are extremely rare. Since Heller et al first described one such case in 1989, only two other cases have been reported to date (Table 1) [1,3,4].

The causes of these extragonadal teratomas have not been well established, but several mechanisms have been proposed, both congenital and acquired. Potential congenital mechanisms for the formation of such a cyst include origination from displaced germ cells and development in a supernumerary ovary. Acquired mechanisms involve autoamputation of an ovarian teratoma secondary to torsion or inflammation and reimplantation with new collateral circulation [6].

In support of a congenital origin, it has been documented that a teratoma can be encountered in diverse organs that coincide with the migration of germ cells during the embryonic period. Furthermore, several authors described ommental teratomas, a type of parasitic teratoma occurring in neonates, as being caused by a supernumerary mechanism [7].

The autoamputation mechanism is especially attractive in the case of a unilaterally absent ovary [1,8]. One reporter described a patient who suffered from abdominal pain and had a mature cystic teratoma with viable ovarian tissue connected by ommental vessels into the cul-de-sac and the absence of an ipsilateral ovary [8]. In the present case, autoamputation of an ovary may be less considered as an explanation of extragonadal teratoma because there was no ovarian tissue in the mass.

Table 1
Review of mature cystic teratomas of the uterosacral ligament.

Case no.	Age (years)	Symptom	MCT location on uterosacral ligament	Ovarian findings	Reference
1	56	Lower back pain	Right	Right ovary: PSCAF Left ovary: PSCAF	4
2	49	Incidental detection	Right	Right ovary: normal Left ovary: normal	3
3	59	Incidental detection	Right	Right ovary: MCT Left ovary: absent	1
4	34	Incidental detection	Left	Right ovary: normal Left ovary: streak	Current case

MCT = mature cystic teratoma; PSCAF = papillary serous cystadenofibroma.

According to our review of the literature, mature cystic teratomas of the uterosacral ligament tend to occur in older patients rather than adolescents, to be located on the right side more frequently than the left side of the uterosacral ligament, and to be detected incidentally (Table 1). In the first reported case, a laparotomy was performed after diagnostic laparoscopy. The other two reported teratomas of this type were removed by conventional laparoscopic surgery. Most recently, we successfully applied LESS making this the first report of a teratoma of the uterosacral ligament to be treated with LESS.

Recently, considerable literatures suggested that LESS correlates with more potential advantages than conventional laparoscopic surgery, including reduced risk of wound infection, hematoma formation, subcutaneous emphysema, and bowel herniation resulting from fewer trocar incisions [9]. Above all, there is general consensus that LESS is superior to conventional laparoscopic surgery in involving better cosmetic outcome due to nearly hidden scar. Moreover, adnexal tumor such as ovarian teratoma is a suitable target for LESS because the uterine manipulator through the vagina provides adequate exposure of the adnexa [9].

There are several disadvantages to LESS including a difficulty in surgical technique and a need for advanced new instruments. In addition, several diseases that lead to dense adhesion or hard mass are considered as an inappropriate indication for LESS because it is difficult to deliver power to the tip of the flexible instrument [10]. In the cases of teratomas of the uterosacral ligament, all tumors were attached to the uterosacral ligament with various intensities of adhesion. However, because they had no distinct ligamentous or vascular connection to other organs, we believe that the application of LESS is acceptable for these tumors in general.

The lack of knowledge regarding extragonadal teratomas may cause surgeons to provide insufficient information to their patients. Therefore, further reports on such rare tumors should continue to be collected.

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