EJGO European Journal of Gynaecological Oncology

Mature cystic teratoma of the vesicovaginal space as an incidental finding on laparoscopy: a case report

Keun Ho Lee, Yong Seok Lee, Eun Kyung Park

Department of Obstetrics and Gynecology, College of Medicine, The Catholic University of Korea, Seoul (Republic of Korea)

Summary

Background: Mature cystic teratomas are the most frequent ovarian tumors; however, teratomas of extragonadal origin are extremely rare. The authors describe herein the first case simultaneously demonstrating normal ovaries and a mature cystic teratoma in the vesicovaginal space on laparoscopy. *Case Report:* A 23-year-old Korean nulliparous woman, with no disease history, was referred to the present outpatient clinic for an asymptomatic pelvic mass detected on routine checkup following a traffic accident. She underwent laparoscopy under a preoperative diagnosis of subserosal myoma in the left anterior wall of the uterus. A benign cystic tumor was found in the vesicovaginal space and was completely treated via laparoscopic resection. *Conclusion:* A mature cystic teratoma in the vesicovaginal space is extremely rare, and proper preoperative diagnosis of this lesion is difficult because of non-specific signs and symptoms. Although uncommon, teratomas should be considered in the differential diagnosis of extragonadal pelvic masses, especially in young adults.

Key words: Cyst; Laparoscopy; Pelvic neoplasms; Teratoma; Vesicovaginal space.

Introduction

Mature cystic teratomas, often referred to as dermoid cysts, are the most common germ cell tumors of the ovary in women of reproductive age. Teratomas are neoplasms of unknown origin composed of tissues resembling normal derivatives of the three germ layers. Although these tumors most commonly occur in the ovaries, they have been documented in a number of other locations (0.4% of all teratomas) [1]. The most common extragonadal site of these teratomas is the omentum [2]. Most teratomas are considered a consequence of autoamputation resulting from torsion with subsequent reimplantation in the abdominal cavity; this is because, in these cases, one of the ovaries was small or absent, or the teratomas contained ovarian tissues [3-5]. Other hypotheses explaining extragonadal teratomas include that they originate from an ectopic ovary or from displaced primordial germ cells. Specifically, teratomas of the vesicovaginal space are even rarer. The present authors report the first case of a mature cystic teratoma that was incidentally found in the vesicovaginal space on laparoscopy in a young adult.

Case Report

This case was approved by the Institutional Review Board of the Catholic University of Korea, Daejeon St. Mary's Hospital. A 23-year-old Korean nullipara woman, with no disease history, was referred to the present outpatient clinic for an asymptomatic pelvic mass detected on routine checkup following a traffic accident. No fever, dysuria, or frequent urination occurred. Her menses was regular, with no history of dysmenorrhea or menorrhagia. No significant medical or surgical history existed. On physical examination, a fixed soft mass was palpated between the left anterior uterus, upper vagina, and bladder. The results on hematological and biochemical investigations and chest radiography were normal. The CA 125 and CA 19-9 levels were within normal limits. Transvaginal ultrasonography showed a round and well-demarcated heteroechogenic mass measuring 6 cm on the lower anterior body of the uterus, without significant vascularity (Figure 1). Moreover, contrast-enhanced computed tomography indicated a 5.7×3.9×5.3-cm round mass in the left anterior pelvic cavity (Figure 2). Accordingly, the primary diagnosis considered was myoma, and a laparoscopic myomectomy was planned.

A three-port laparoscopy (10-mm subumbilical and 5-mm suprapubic ports on either side) was performed, using a routine open entry approach. On inspection of the abdominal cavity, there was no mass-like lesion. The uterus, both ovaries, and the uterine tube appeared normal (Figure 3). The mass was deep inside the left vesicovaginal space between the vagina and under the uterine cervix and bladder. An incision was made at the peritoneum above the mass, underlining the mass formation. The operation was continued with careful dissection of the cystic wall from the neighboring structures by using a monopolar hook dissector and harmonic scalpel. The vesicovaginal space under the insertion site of the left ureter was dissected, and the dilated left ureter was revealed. The cyst was ruptured, and cheese-like material containing short fine hairs was drained (Figure 4). The remainder of the lesion was dissected from the surrounding pelvic tissue. The mass had no feeding vessel or ligamentous connection to other organs

Revised manuscript accepted for publication October 12, 2016

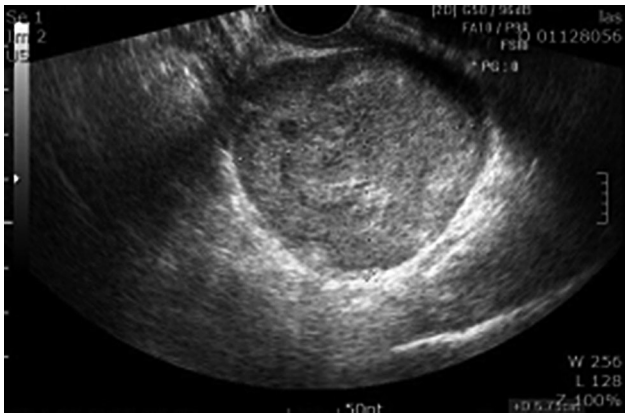

Figure 1. — Transvaginal ultrasonography showing a solid mass on the left lower anterior body of the uterus.

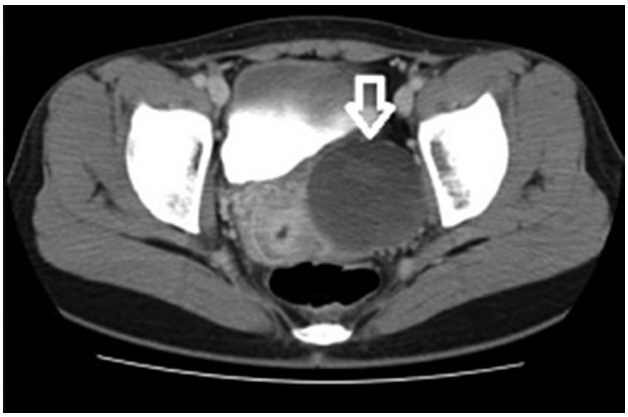

Figure 2. — Contrast-enhanced computed tomography indicated a round mass in the left anterior pelvic cavity.

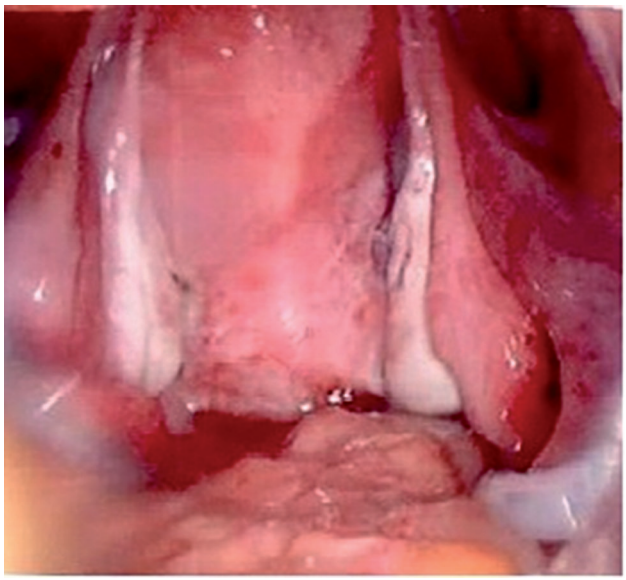

Figure 3. — Both ovaries and the uterine tube are normal

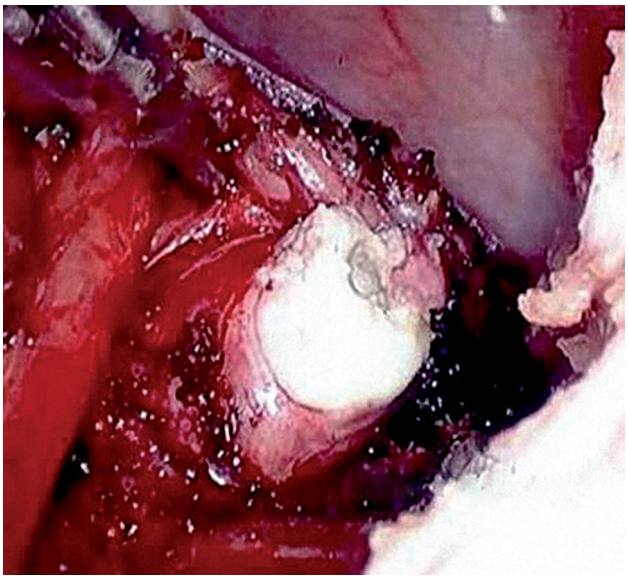

Figure 4. — Dilated ureter on the left side and ruptured teratoma in the vesicovaginal space.

except for the attachment of fine vessels to the upper vagina and bladder. The total mass was removed laparoscopically. The fully dissected cyst was placed in an Endobag and removed through an umbilical incision. The urinary catheter was removed on postoperative day 1, and the patient recovered uneventfully and was discharged two days after the surgery. Microscopically, the cyst contained predominantly keratinous material with some squamous epithelial cell lining and bundles of smooth muscle. No immature elements were identified, and there was no evidence of malignancy. The pathological diagnosis confirmed a mature cystic teratoma.

Discussion

Mature cystic teratomas occur most commonly in the ovary, accounting for approximately 20% of all adult ovarian neoplasms [6]. Extragonadal teratomas occur anywhere along the midline of the body, such as the mediastinum, because of the migration of germ cells in embryonic life. However, extragonadal teratomas in the abdominal cavity are extremely rare, and the most common extragonadal site is the omentum, where 32 cases in total have been reported [7]. To the present authors' knowledge, this is the first re-

ported case simultaneously displaying normal ovaries and a teratoma in the vesicovaginal space on laparoscopy.

The causes of extragonadal teratomas have been poorly understood, but several mechanisms have been proposed. It is generally known that teratomas arise from germ cells originating in mature gonads. Migration of germ cells from the yolk sac along the hindgut (route of mesentery) toward the genital ridge (primitive gonad) takes place during early fetal development. These totipotent cells (or primordial germ cells or early embryonic cells) may give rise to a variety of tissues originating from the three primitive embryonic layers. Three proposed theories for extragonadal sites of teratoma exist: [1] autoamputation of an ovarian teratoma and reimplantation into an extragonadal site, [2] development in a supernumerary ovary, and [3] origination from displaced germ cells [8].

The first theory is that extragonadal teratomas may be autoamputated into another site of the abdominal cavity from an ovarian site. In relation to this, torsion of the pedicle is reported to be the most frequent complication of ovarian teratomas, occurring in 16.1% of cases [2]. Torsion interferes with the blood supply of the involved organ, and venous congestion and aseptic inflammation of the tumor wall may thus result. In acute torsion, the tumor undergoes necrosis and subsequent atrophy because of ischemia. In subacute or chronic torsion, the tumor may adhere to adjacent structures with the formation of new collateral circulation.

The omentum, because of its special role in intra-abdominal inflammation defense processes, is probably the main location for secondary implantation of the tumor. When one of the ovaries is small or absent or when teratomas contain ovarian tissues, autoamputation has been thought to be a possible cause. This theory is widely accepted as the etiology of extragonadal teratomas in the abdominal cavity. However, in the present case, the teratoma contained no ovarian tissues, and both ovaries were intact on laparoscopy. Thus, autoamputation may not be a reasonable theory in the present case.

There is also lack of support for the second and third theories, as no reported cases are documented in the published literature. The second theory is that extragonadal teratomas may occur in an ectopic ovary [5], which is thought to arise congenitally or following surgery or pelvic inflammation. The present patient had no history of laparotomy, and the tumor contained no ovarian tissues. Therefore, this theory does not appear to be applicable.

The third theory posits that extragonadal teratomas may originate from displaced primordial germ cells. Primordial germ cells may stop differentiating during migration, thus later causing a teratoma [9]. The present authors believe this theory is most applicable to the current case, although it is difficult to prove. During migration to the genital ridge, DNA methylation of primordial germ cells is erased and subsequently reestablished during gametogenesis [10]. The results of genomic imprinting research have indicated that each group of germ cell tumors has a different imprinting status [11]. For example, teratoma/yolk sac tumors in infants and teratomas of the ovary in adults have a different imprinting status. Accordingly, further investigation into the imprinting status in the teratoma may have been helpful in understanding the origin of the tumor.

It is difficult to establish a preoperative diagnosis in cases such as the present one, although ultrasonography, computed tomography, and magnetic resonance imaging may aid in the diagnosis. The treatment of choice for retroperitoneal tumors is complete surgical excision, since definitive diagnosis is only achieved following histopathological examination of the specimen. The present case caused confusion to the surgeon, as there was no pelvic mass in the abdominal cavity on laparoscopy. Based on the location of the mass from previous pelvic examination, transvaginal ultrasonography, and computed tomography, the present authors dissected the left anterior peritoneum of the vesicocervical space and discovered the mass. Complete excision of the mass was achieved via laparoscopy.

In conclusion, because the specimen of the current case had no ovarian tissues and both ovaries were normal, the teratoma was considered to have originated primarily in the vesicovaginal space. Retroperitoneal teratomas of the vesicovaginal space are rare and difficult to diagnose early owing to non-specific signs and symptoms; however, they should be considered in the differential diagnosis of a pelvic mass in young adults. Once the diagnosis is made, surgical removal is necessary because of the uncertain course of the disease.

References

[1] Koo Y.J., Im K.S., Jung H.J., Kwon Y.S.: "Mature cystic teratoma of the uterosacral ligament successfully treated with laparoendoscopic single-site surgery". *Taiwan J. Obstet. Gynecol.*, 2012, *51*, 86.

[2] Peterson W.F., Prevost E.C., Edmunds F.T., Hundley J.M., Morris F.K.: "Benign cystic teratomas of the ovary; a clinico-statistical study of 1,007 cases with a review of the literature". *Am. J. Obstet. Gynecol.*, 1955, *70*, 368.

[3] Khoo C.K., Chua I., Siow I., and Chern B.: " Parasitic dermoid cyst of the pouch of Douglas: a case report". *J. Minim. Invasive Gynecol.*, 2008, *15*, 761.

[4] Ohshima K., Umeda A., Hosoi A., Yamamoto T., Munakata S.: " Mature cystic teratoma in Douglas' pouch". *Case Rep. Pathol.*, 2015, *2015*, 202853.

[5] Sinha R., Sundaram M., Lakhotia S.: " Multiple intraabdominal parasitic cystic teratomas". *J. Minim. Invasive Gynecol.*, 2009, *16*, 789.

[6] Talerman A., Vang R.: "Germ cell tumors of the ovary". *In:* Kurman R.J., Ellenson L.H., Ronnett B.M., (eds). *Blaustein's Pathology of the Female Genital Tract.* New York: Springer; 2008, 967.

[7] Hegde P.: "Extragonadal omental teratoma: a case report". *J Obstet Gynaecol Res.*, 2014, *40*, 618.

[8] Ushakov F.B., Meirow D., Prus D., Libson E., Benshushan A., Rojansky N.: "Parasitic ovarian dermoid tumor of the omentum-A review of the literature and report of two new cases". *Eur. J. Obstet. Gynecol. Reprod. Biol.*, 1998, *81*, 77.

[9] Oosterhuis J.W., Stoop H., Honecker F., Looijenga L.H.: "Why

human extragonadal germ cell tumours occur in the midline of the body: old concepts, new perspectives". *Int. J. Androl.*, 2007, *30*, 256.
[10] Bartolomei MS, Ferguson-Smith AC.: "Mammalian genomic imprinting". *Cold Spring Harb Perspect Biol.*, 2011, *3*, a002592.
[11] Sievers S, Alemazkour K, Zahn S, Perlman EJ, Gillis AJ, Looijenga LH, *et al.*: "IGF2/H19 imprinting analysis of human germ cell tumors (GCTs) using the methylation-sensitive single-nucleotide primer extension method reflects the origin of GCTs in different stages of primordial germ cell development". *Genes Chromosomes Cancer, 2005, 44, 256.*

Corresponding Author:
EUN KYUNG PARK, M.D, PHD
Department of Obstetrics and Gynecology
The Catholic University of Korea
222 Banpo-daero Seocho-gu
Seoul 06591 (Republic of Korea)
e-mail: guevara614@catholic.ac.kr