

Mature Cystic Teratoma of the Uterine Surface and Ovary with Adenocarcinoma of the Endometrium: An Unusual Case Scenario and Literature Review

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Abstract

Teratomas that occur in the uterus are exceedingly rare. To the best of our knowledge there are only 22 cases of mature and immature teratomas of the uterus and cervix thus far reported in the literature. We report an unusual case of mature cystic teratoma of the uterine surface with well-differentiated adenocarcinoma of the endometrium and uterine leiomyoma along with a mature cystic teratoma of the right ovary.

Keywords: Mature cystic teratoma, Uterus and ovary

Introduction

Teratomas are the most common germ cell tumors composed of multiple cell types derived from one or more of the three embryonic germ cell layers: ectoderm, endoderm, and mesoderm.¹

Teratomas may be classified as mature or immature on the basis of the presence of immature/embryonic elements.^{1,2} They usually arise in the gonads and often occur in infancy and childhood. Extragonadal teratomas are rare and mainly develop in midline structures. Common sites are the retroperitoneum and mediastinum.¹

Occurrence of teratoma in the

uterus is exceedingly rare.² Mann in 1929 was the first to describe a case of primary mature teratoma in the uterus.³ To the best of our knowledge, since then only 22 cases of mature and immature teratomas of the uterus and cervix have been reported.

We report a case of mature cystic teratoma of the uterine surface with well-differentiated adenocarcinoma of the endometrium and uterine leiomyoma along with a mature cystic teratoma of the right ovary.

Case Report

A 62-year-old postmenopausal woman, para 3 gravida 3, presented to our hospital with a history of

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Figure 1. Right ovarian mass.

vaginal bleeding and whitish discharge since 2 months. She was a known case of diabetes and hypertension on medications. Ultrasound of the abdomen and pelvis showed a bulky uterus with abnormally thickened and echogenic endometrium with subserosal uterine fibroids, a normal appearing left ovary and 11×10×9 cm right adnexal mass suspicious for cystadenocarcinoma. Magnetic resonance imaging (MRI) showed a bulky, lobulated uterus with large lesions on the anterior, lateral, and posterior walls which were hypointense on T1 and T2 weighted images. There was a 2.2 cm thickened endometrium and complex right adnexal mass (11.8×7.1×5.8 cm) which had solid and cystic areas with inner areas of T1 weighted hyperintensity suppressing on



Figure 3. Bulky uterus with sessile mass at the fundal surface with cervix.

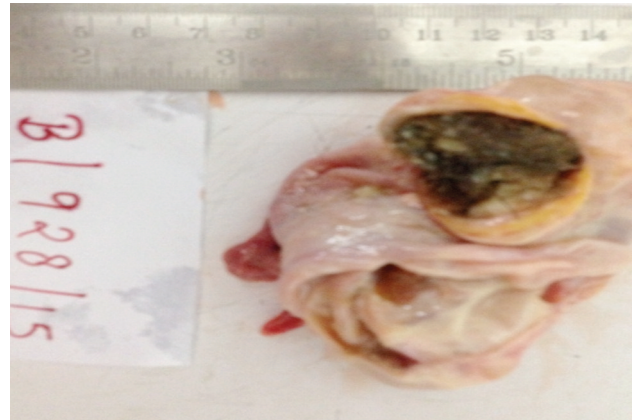


Figure 2. Cut surface of the right ovarian mass with cystic and solid areas that show a tuft of hair.

infrared images of questionable mitotic etiology with blood/fat content within. Serum tumor markers, alpha-fetoprotein (AFP), and human choriongonadotropin (HCG) were within normal limits. However, lactate dehydrogenase (LDH) and thyroid stimulating hormone (TSH) were marginally elevated. The patient underwent an exploratory laparotomy with right ovarian mass excision, frozen section, and total abdominal hysterectomy. The postoperative period was uneventful.

Pathological findings were as follows. A right ovarian mass (12.4×9.2×7.0 cm) with attached fallopian tube was received for intraoperative consultation. The external surface was bosselated, smooth, and glistening. The cut section revealed multiple solid and cystic areas filled with pultaceous material, a tuft of hair and solid areas of variegated appearance (Figures 1, 2).

The bulky uterus with cervical specimen



Figure 4. Cut surface of the sessile mass at the uterine surface that shows teeth and a tuft of hair.



Figure 5. Cut surface of the uterus with endometrial polypoidal growth and an intramural fibroid.

measured 12.5×7.4×6.6 cm. The external surface of the uterus was bosselated with a large sessile mass at the fundus that measured 3.5×3.5×3.0 cm (Figure 3). The cut surface of the sessile mass had multiple solid and cystic areas filled with pultaceous material, a tuft of hair, and five teeth (Figure 4). The cut surface of the uterus had a polypoidal growth that measured 5.0×2.0×2.0 cm which arose from the distorted endometrial cavity up to the upper end of the isthmus. Also present was an intramural fibroid that measured 6.0 cm in maximum diameter and a subserosal fibroid that measured 2.0 cm in maximum diameter (Figure 5). The cervix was unremarkable.

Microscopic examination showed a mature cystic teratoma of the right ovary and uterus. The tumor was composed of mature elements of all three germ cell layers with thyroid follicles,

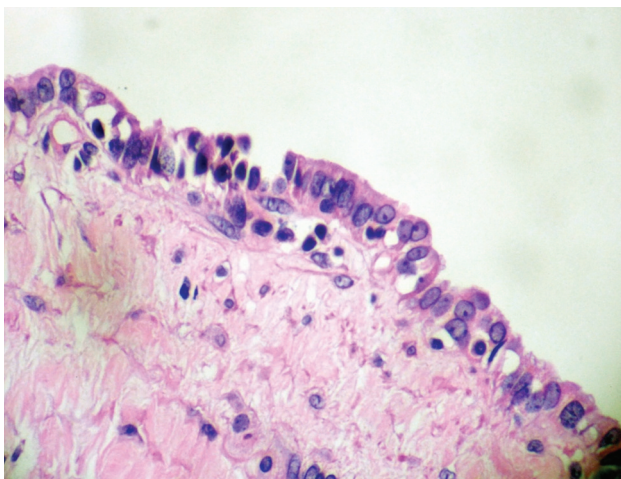


Figure 7. Photomicrograph of the sessile mass at the uterine surface that shows respiratory epithelium (H&E, 400×).

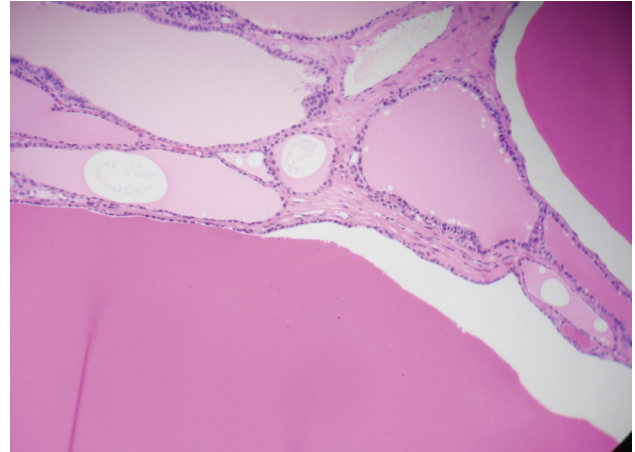


Figure 6. Photomicrograph of the sessile mass at the uterine surface that shows thyroid follicles (H&E, 400×).

squamous epithelium, hair follicles, respiratory columnar epithelium, sebaceous glands, smooth muscle, adipose tissue, microcalcification, teeth, bone, a number of blood vessels, and lymphatics (Figures 6-12). No immature elements or infiltration into the myometrium were observed. Sections from the endometrial polypoidal growth showed a well-differentiated adenocarcinoma of the endometrium that invaded less than half of the adjacent myometrium (Figure 13). Sections from the myometrium showed an intramural and a subserosal leiomyoma. However, right and left fallopian tube as well as isthmus and cervix were unremarkable. The peritoneal wash fluid was negative for malignant cells.

Discussion

The uterus is a rather rare site for the

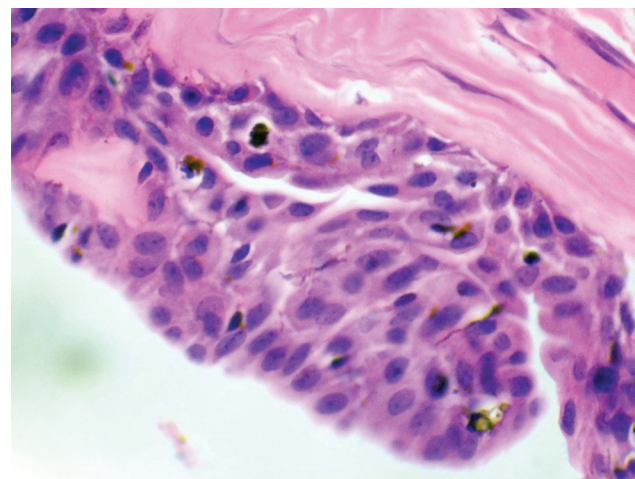


Figure 8. Photomicrograph of the sessile mass at the uterine surface that shows squamous epithelium (H&E, 400×).

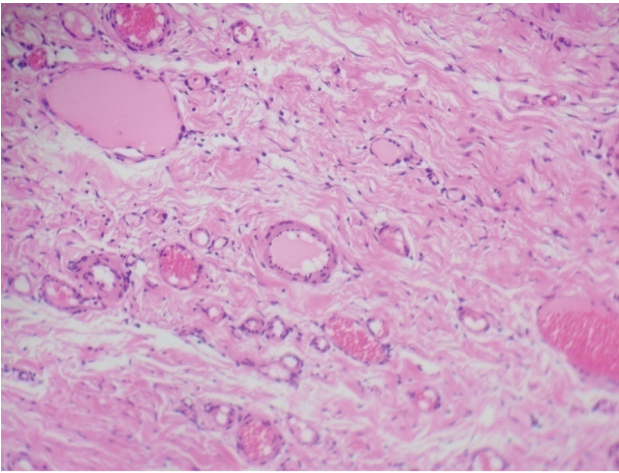


Figure 9. Photomicrograph of the sessile mass at the uterine surface with numerous (smaller and larger) blood vessels and lymphatics (H&E, 400×).

development of a primary teratoma. In a literature review by Iwanaga et al. in 1993, there were a total of 15 cases reported, which included their own case.² Subsequently 7 more cases have been reported.^{1, 4-9} Based on a review of these reports, the teratomas either arose from the uterine cavity or from the cervical canal. Until now, only one case of an immature teratoma that arose from the uterine fundus² and only one case with coexistent immature teratoma of the uterine cavity and endometrial adenocarcinoma have been reported.¹⁰ The present case was the only reported case of a mature teratoma that arose from the uterine fundal surface with coexistent mature cystic teratoma of the right ovary, a well-differentiated adenocarcinoma of the endometrium, an intramural, and a subserosal leiomyoma.

Uterine teratomas occur typically in the second

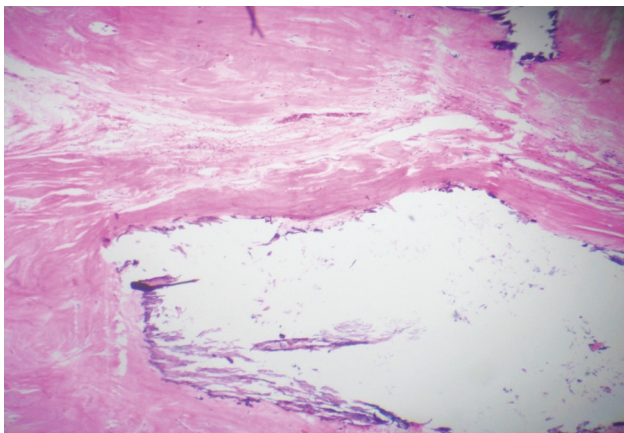


Figure 11. Photomicrograph of sessile mass at the uterine surface that shows microcalcifications (H&E, 400×).

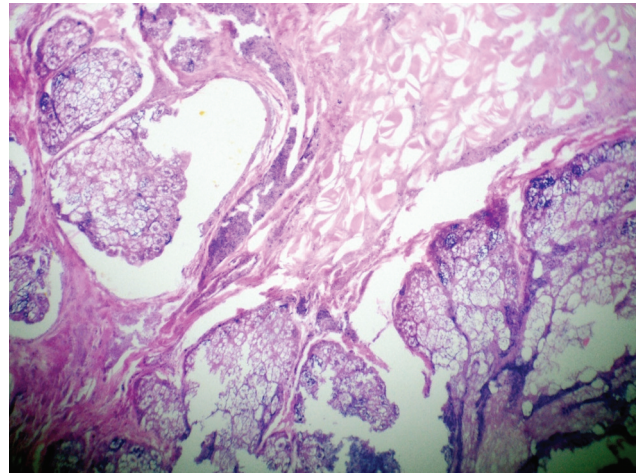


Figure 10. Photomicrograph of the sessile mass at the uterine surface that shows sebaceous glands (H&E, 400×).

to fourth decades of life.² They are seen very rarely in postmenopausal women as seen in our patient.

The histogenesis of an extragonadal teratoma has always attracted interest. It is hypothesized that uterine teratomas originate either from pluripotent embryonic cells (i.e., residual tissues that remain in the uterus after a missed abortion) because the genital canal is the natural pathway for the fertilized ovum (blastomere theory). However this theory has been discredited as teratoma cells have a 46, XX karyotype with identical X chromosomes derived solely from the host.

According to the parthenogenetic theory, teratomas arise from primordial cells that have gone astray during embryogenesis with extragonadal teratoma growth in sites where germ cells normally migrate in early embryonic life.^{1, 2, 8}

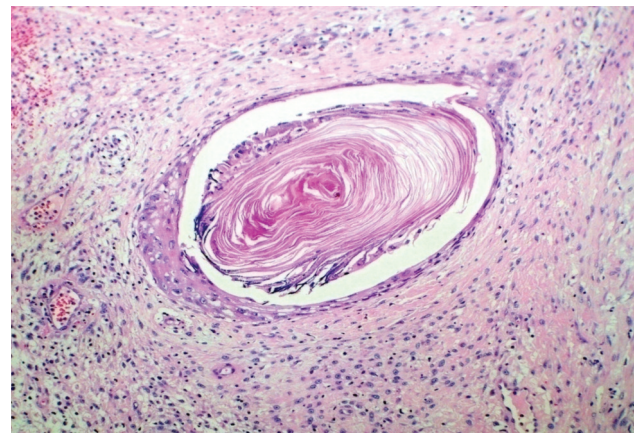


Figure 12. Photomicrograph of a sessile mass at the uterine surface that shows keratin pearl (H&E, 400×).

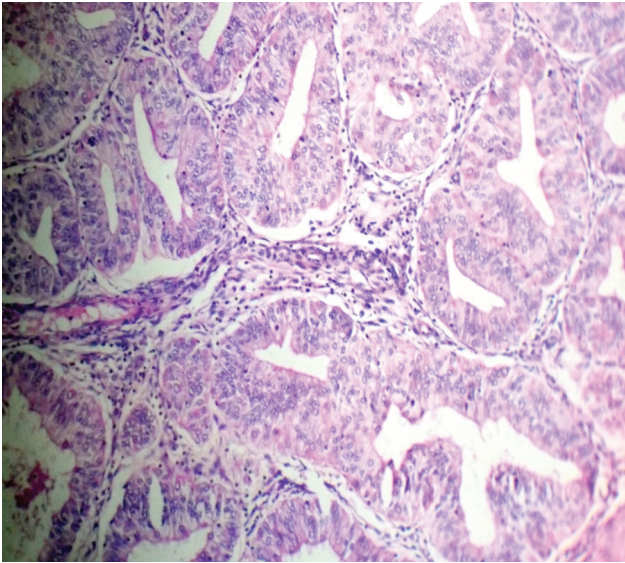


Figure 13. Photomicrograph of the endometrial polypoidal growth that shows a well-differentiated endometrium (H&E, 400×).

However, in the present case, the patient was postmenopausal for six years and the tumor site differed from previously reported cases. In the previous cases the tumors were found in the uterine cavity or the cervical canal, while in the present case the tumor was at the uterine fundal surface. One could speculate that the teratoma in this case might have arisen either from a germ cell which had gone astray during early embryogenesis or from a germ cell which was primarily intraovarian but became displaced into the uterine fundus and arrested.²

Conflict of interest

No conflict of interest is declared.

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