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Case Reports

Coexistent mesenteric and ovarian mature cystic teratomas: a case report

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Summary

This report describes the first documented case of coexistent mesenteric and gonadal teratomas in an adult female patient. Physical examination of a 51-year-old Korean woman referred for treatment of abdominal distension and pain revealed two masses in both the right upper abdomen and in the right pelvic region. Computed tomography (CT) of the abdomen and pelvis showed the presence of well-defined, complex, fat-dense mass lesions in the upper abdomen and pelvic cavity. A large cystic mass located in the retroperitoneal space extending from the mesenteric border at the level of the transverse colon, and a goose-egg sized right ovarian mass were founded on exploratory laparotomy. The entire abdominal tumor was excised and total hysterectomy with bilateral salpingo-oophorectomy was performed. Examination of the macroscopic and microscopic findings led to diagnosis of mature cystic teratomas of the ovary and the mesentery. The patient's postoperative course was uneventful.

Key words: Mature cystic teratoma; Mesentery; Ovary; Coexistent.

Introduction

Mature cystic teratoma is one of the most common forms of ovarian neoplasm. It is a congenital germ cell tumor (GCT) that can contain somatic tissues from all three germ layers. Most teratomas arise in the gonads, but may occasionally occur in extragonadal sites, such as the sacrococcygeal region, mediastinum, neck, and retroperitoneum [1]. All forms of teratomas occur more commonly in children than in adults. Among the forms of teratomas, retroperitoneal teratomas often occur in infancy and childhood but rarely in adulthood. This case report describes the first documented case of coexistent mature cystic teratomas in the right ovary and mesenteric region.

Case Report

A 51-year-old Korean woman referred for treatment of abdominal distension and pain, which she had been experiencing for two years with no concomitant weight loss or bowel or urinary symptoms. Physical examination revealed a fetal-head-sized palpable mass in the right upper abdomen, and pelvic examination revealed a goose-egg-sized right adnexal mass. Further examination revealed no other obvious abnormalities. The results of routine blood testing and urinalysis revealed serum and biochemical parameters within normal limits, including normal levels of tumor markers cancer antigen (CA) 19-9, CA 125, alpha-fetoprotein (AFP), and carcinoembryonic antigen (CEA).

Computed tomography (CT) of the abdomen and pelvis showed the presence of well-defined, complex, fat-dense mass lesions in the upper abdomen and right adnexa, and the absence of lymphadenopathy and ascites (Figure 1). Exploratory laparotomy disclosed a large cystic mass behind the transverse colon located in the retroperitoneal space and extending from the mesenteric border (Figure 2). Based on these findings, the entire abdominal tumor was excised and total hysterectomy with bilateral salpingo-oophorectomy was performed. Macroscopically, the encapsulated abdominal mass measured 9.0 x 8.0 x 6.0 cm and the right adnexal mass 7.2 x 5.5 x 4.5 cm. Microscopically, both masses presented as mature cystic teratomas comprising squamous epithelium, skin adnexa, and fat (Figure 3). Based on analysis of the macroscopic and microscopic findings, the patient was diagnosed with mature cystic teratomas of the ovary and extragonadal region. The patient's postoperative course was uneventful.

Discussion

The designation *teratoma* refers to a varied group of tumors that show differentiation toward somatic-type tissues that can be typical of either adult or embryonic development. Teratomas are the most common type of GCT, and most, but not all, are benign. The component tissues in a teratoma, which can range from immature to well-differentiated tissues, are foreign to the anatomic site in which they are found. Most teratomas are cystic and composed of mature adult-type tissues. Better known

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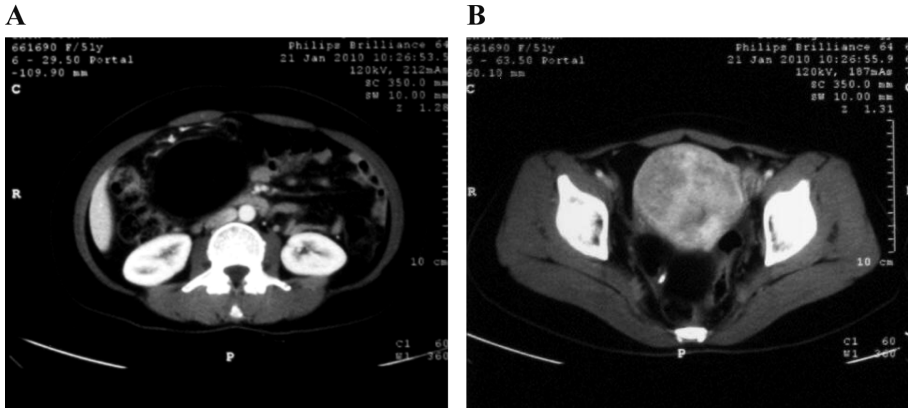

Figure 1. — CT scan of (A) complex fat-dense mass lesion with rounded group of linear isodense elements, possibly elements of a hair-plug in the upper abdomen, and (B) fat- and sheet-like calcifications of the right adnexa.

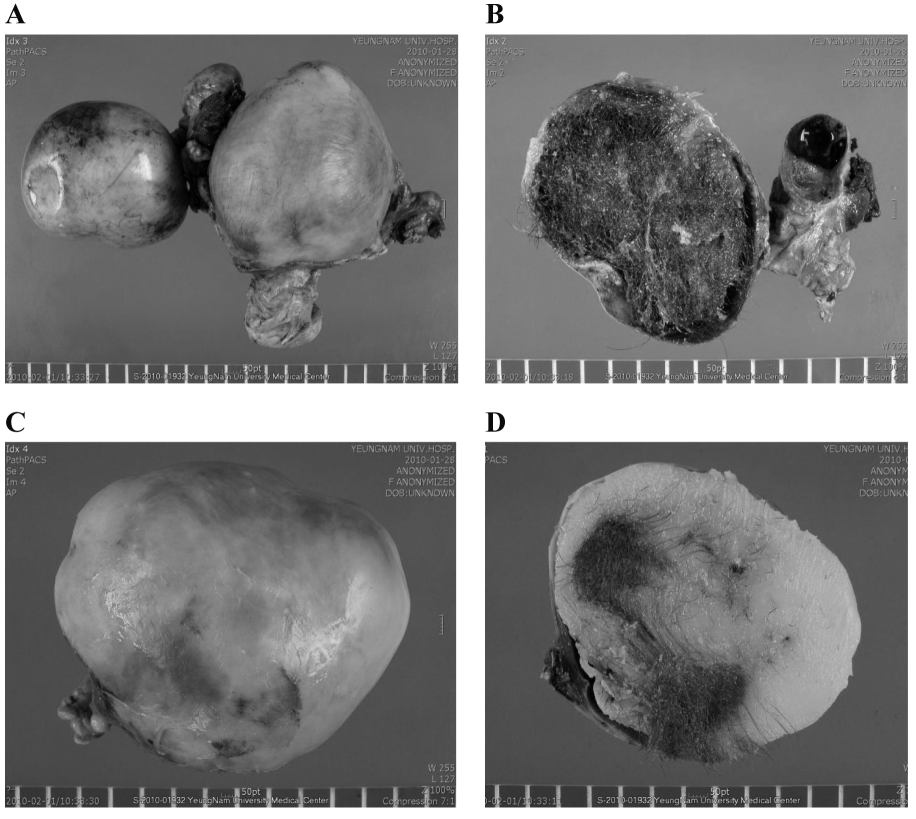

Figure 2. — Gross appearance of teratomas. View of (A) enlarged uterus and right adenxa, (B) cut section of right adenxa, (C) upper abdominal mass attached to mesenteric border of transverse colon, and (D) cut section of upper abdominal mass.

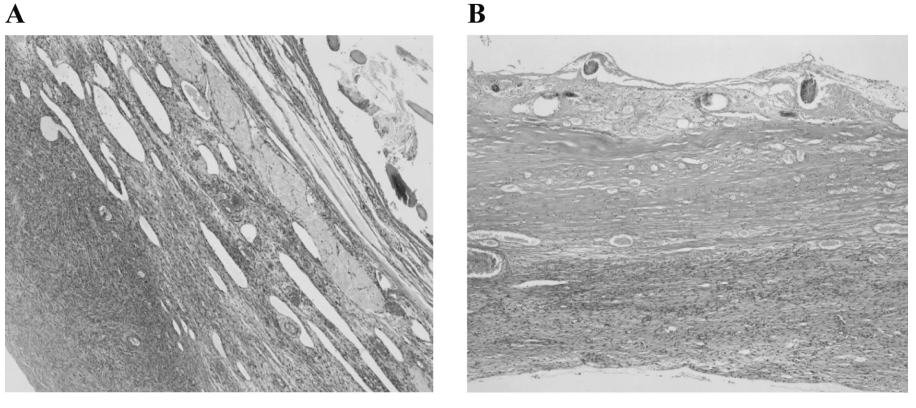

Figure 3. — Microscopic view of teratomas. (A) View of right ovary showing a small mature cystic teratoma containing hair shafts (upper right) in the lumen and ovarian cortex (lower left) and corpus albicans (middle) in the wall (×40, H&E staining). (B) View of tumor of mesenteric border appearing as a large mature cystic teratoma containing bundles of degenerated hair shafts. Ovarian stromal cell-like spindle cells can be observed in the wall. These findings indicate a high possibility that the tumor originated from extragonadal germ cells (×40, H&E staining).

as dermoid cysts, these mature cystic teratomas are the most common form of ovarian tumor in women in the second and third decade of life, accounting for more than 95% of all ovarian teratomas, almost all of which are benign [2].

The most widely accepted theory of germ cell development proposes that teratomas arise from totipotential germ cells that fail to mature normally in the gonadal regions. These totipotential cells can differentiate into tissue components of the mesoderm, ectoderm, and endoderm. GCTs that arise outside the gonadal regions are classified as extragonadal GCTs if there is no evidence of primary tumor in the testes or ovaries. Totipotential germ cells tend to be migratory, which may account for the development of gonadal and extragonadal teratoma of the midline structure, with the specific site varying with age. In adults, the most common extragonadal GCT sites are, in order of frequency, the anterior mediastinum, retroperitoneum, and the pineal and suprasellar regions. In infants and young children, the most common sites are the sacrococcygeal and intracranial regions. The overall incidence of all forms of teratomas is higher in children than adults. Teratomas have been found to arise in various locations in children, including (in order of frequency) the ovaries, the testes, the anterior mediastinum, the retroperitoneal space, and the sacrococcygeal region [1].

Primary retroperitoneal extragonadal teratomas, which are often found near the upper pole of left kidney [3], account for 1–11% of all primary retroperitoneal tumors [2]. Retroperitoneal teratomas have been misdiagnosed as ovarian and adrenal tumors, including adrenal myelolipomas, Wilms' tumors, renal cysts, retroperitoneal fibromas, sarcomas, hemangiomas, and enlarged lymph nodes [4, 5]. Primary retroperitoneal teratomas are rare in adults and more common in children and young adults; their incidence is almost equal in both genders [3]. Benign teratomas are usually asymptomatic and, as such, may be diagnosed incidentally. However, as the tumor mass increases, the surrounding structures may be compressed, leading patients to experience obstructive symptoms, such as abdominal distension, pain, nausea, and vomiting [6]. Physical examination to identify the source of these symptoms may lead to detection of an abdominal mass. Malignant teratomas are often diagnosed at advanced stages and, as such, tend to progress rapidly [3]. The malignancy potency of retroperitoneal teratoma is significantly higher in adults than children, with the malignancy incidence 25.8% and 6.8%, respectively [7]. However, 24% of neonatal retroperitoneal teratomas are malignant [8].

Plain abdominal radiography of retroperitoneal teratoma cases always reveals a soft tissue mass. Calcifications within the tumor or on the rim of the cyst wall or teeth and bones appear in 61.5% of retroperitoneal teratoma cases, and are thus useful in preoperative diagnoses [2]. However, as calcifications also occur in 12.5% of malignant teratomas, they are not indicators of benign teratomas [9].

Among the means of examination and evaluation, ultrasonography provides specific data regarding the cystic, solid, or complex components of a teratoma; the acoustic shadow induced by calcifications in the teratoma; and, occasionally, fat-fluid levels in the teratoma. CT is a more accurate diagnostic tool than ultrasonography because it provides data that help in defining the extent to which the teratoma affects surrounding organs and in evaluating the specific components [10]. Magnetic resonance imaging (MRI) provides data with which to determine the anatomical relationship between the abdominal aorta or spinal cord and the spread and extent of the local tumor, facilitating prediction of resectability, and evaluation of recurrence [11]. Regardless of the means of examination, definitive diagnosis can only be made after histological analysis, and surgery is paramount in both diagnosis and treatment.

The primary treatment of retroperitoneal teratoma is surgical resectioning. In adults, primary retroperitoneal teratomas typically do not invade surrounding structures, and can be completely resected [4]. Prognosis depends on the tissue components of the teratoma. The prognosis is excellent for benign retroperitoneal teratomas that can be completely resected [12]. However, as malignancy usually recurs in spite of surgery, the prognosis for malignant retroperitoneal teratoma is poor, with a median survival of 18 months among malignant teratoma patients [9].

This report describes the first documented case of mature cystic teratoma, a rare condition, arising in the extragonadal (mesenteric) region and the right ovary of a 51-year-old woman. After CT revealed coexistent ovarian and extragonadal teratomas showing typical morphology, the tumor was resected. Subsequent histopathological examination confirmed a diagnosis of coexistent primary benign mesenteric and ovarian teratomas. The patient experienced an unremarkable postoperative recovery.

References

[1] Yoon S.S., Tanabe K.K., Warshaw A.L.: "Adult primary retroperitoneal teratoma". *Surgery*, 2005, *137*, 663.

[2] Ayhan A., Bukulmez O., Genc C., Karamursel B.S., Ayhan A.: "Mature cystic teratomas of the ovary: case series from one institution over 34 years". *Eur. J. Obstet. Gynecol. Reprod. Biol.*, 2000, *88*, 153.

[3] Gatcombe H.G., Assikis V., Kooby D., Johnstone P.A.: "Primary retroperitoneal teratomas: a review of the literature". *J. Surg. Oncol.*, 2004, *86*, 107.

[4] Yumura Y., Chiba K., Urushibara M., Saito K, Hirokawa M.: "A case of retroperitoneal teratoma difficult to distinguish from adrenal myelolipoma". *Hinyokika Kiyo*, 2000, *46*, 891. [Article in Japanese]

[5] Wang RM, Chen CA. Primary retroperitoneal teratoma. Acta Obstet Gynecol Scand 2000; 79: 707–708.

[6] Gschwend J., Burke T.W., Woodward J.E., Heller P.B.: :Retro-peritoneal teratoma presenting as an abdominal-pelvic mass". *Obstet. Gynecol.*, 1987, *70*, 500.

[7] Lambrianides A.L., Walker M.M., Rosin R.D.: "Primary retroperitoneal teratoma in adults". *Urology*, 1987, *29*, 310.
[8] Augé B., Satgé D., Sauvage P., Lutz P., Chenard M.P., Levy J.M.: Retroperitoneal teratomas in the perinatal period. Review of the literature concerning a neonatal, immature, aggressive teratoma". *Ann. Pediatr. (Paris)*, 1993, *40*, 613.
[9] Bruneton J.N., Diard F., Drouillard J.P., Sabatier J.C., Tavernier J.F.: "Primary retroperitoneal teratoma in adults: presentation of two cases and review of the literature". *Radiology*, 1980, *134*, 613.
[10] Davidson A.J., Hartman D.S., Goldman S.M.: "Mature teratoma of the retroperitoneum: radiologic, pathologic, and clinical correlation". *Radiology*, 1989, *172*, 421.
[11] Cohen M.C., Weetman R.M., Provisor A.J.: "Efficacy of magnetic resonance imaging in 139 children with tumors". *Arch. Surg.*, 1986, *121*, 522.
[12] Pantoja E., Llobet R., Gonzalez-Flores B.: "Retroperitoneal teratoma: a historical review". *J. Urol.*, 1976, *115*, 520.

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