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Case report

Malignant melanoma arising in a mature teratoma: A case report with review of the recent literature

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ABSTRACT

Mature cystic teratomas constitute 10–20% of all ovarian neoplasms. Malignant transformation is very rare occurring in only 0.1–2% of mature teratoma cases. Malignant melanoma is among the least common transformations. Herein, we describe a case of young woman initially undergoing evaluation for infertility who was found to have malignant melanoma arising in a mature dermoid cyst. She subsequently underwent unilateral salpingo-oophorectomy with staging procedure with benign pathology. There was no need for adjuvant therapy and the patient is without disease to date (nearly 10-months in follow-up now). We reviewed the existing literature and this is one of only a few cases documented in the last decade.

1. Introduction

Although mature teratomas (MTs), also known as dermoid cysts, compromise 10–20% of ovarian neoplasms globally (Park et al., 2008), malignant transformation is very rare occurring in only 0.17–2% of MT cases (Hackethal et al., 2008). The most frequent malignancy arising in MTs is squamous cell carcinoma (88.3%) (Crouet et al., 1986), followed by adenocarcinoma, fibrosarcoma, carcinoid tumor and mixed tumors. Malignant melanoma is among the least common transformations, with estimated incidence of 0.2–0.8% (Mandal et al., 2010).

A review of 36 cases during 1901 to 2009 has been reported by Xu et al. (2011), showing only six patients surviving >2-years, while only one patient had no evidence of disease (NED) 5-years post-diagnosis. Another review by Ueng et al. (2010) discussed four cases of this tumor type. Prognosis was poor, with all dying 3, 17, 18 and 5 months, respectively, post-diagnosis. Herein, we describe a rare case of this tumor type and discuss various options for management/treatment.

2. Case presentation

A 35-year-old Hispanic female, G0, referred by her Reproductive Endocrinologist. While undergoing evaluation for infertility, pelvic ultrasound demonstrated a pelvic mass versus fibroid measuring 6.2 × 6.6 cm, midline to the left. The left ovary contained multiple follicles measuring 3.4 × 1.8 cm, and a dominant follicle measuring 2.4 × 1.8 cm. There was a contiguous mass, which measured 8.2 × 6.3 × 7.4 cm, present anteriorly, abutting the uterus and left ovary. The lesion was suggestive of a large hemorrhagic cyst or an endometrioma. She complained of pain in left lower quadrant that was crampy in nature.

The patient underwent surgical assessment in conjunction with Reproductive Endocrinology. Diagnostic hysteroscopy followed by laparoscopy was performed revealing a 10 cm mass apparently originating from right ovary and two pedunculated uterine fibroids, which measured ~2 cm and ~5 mm, respectively. Given the size of ovarian mass, the procedure was converted to laparotomy and the ovary was exteriorized. The right ovary was exteriorized from the abdominal cavity. During cystectomy, the cyst ruptured without evidence of intra-abdominal spillage. A myomectomy was also performed. Pathology revealed a unilocular cyst filled with sebaceous material and hair. A 1-cm Rokitansky nodule was present but no other nodules or papillations were identified. Microscopic examination of the ovarian cyst revealed a mature cystic teratoma containing 7 mm focus of melanoma, which was predominantly intra-epidermal; but contained a few

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small nests of invasive melanoma in the adjacent stroma, associated with a dense lymphocytic infiltrate (Figs. 1–3). No lymphatic invasion was identified and the melanoma did not involve surface of the ovary. Histologic sections were reviewed at Johns Hopkins Pathology Department with diagnostic concurrence.

For further assessment, CT scan was performed post-operatively. This showed a complex left ovarian cyst with a thickened wall, measuring 2.3 cm and a simple-appearing right ovarian cyst measuring 3.1 cm, and showed no evidence of retroperitoneal lymphadenopathy. PET scan was also obtained which showed no evidence of metastasis. The case was reviewed at our multi-disciplinary tumor board. Given the pathological findings of the malignant melanoma on initial surgery, recommendation was to perform right salpingo-oophorectomy and a staging procedure.

The patient then underwent robotic-assisted laparoscopic right salpingo-oophorectomy, right pelvic and aortic lymphadenectomy, omental biopsy, intra-peritoneal biopsies and left ovarian cystectomy. All pathology was benign. The right tube and ovary showed no evidence of residual melanoma.

Post-operatively, the patient did well and is without disease to date (nearly 10-months in follow-up now). The case was again discussed at tumor board and no further treatment was recommended, other than close surveillance.

3. Discussion

Mature cystic teratomas (MCTs) constitute 10–20% of all ovarian neoplasms. They tend to present in young women, around the age of 30-years. MCTs are composed of well-differentiated derivations from at least two of the three germ cell layers (i.e., ectoderm, mesoderm, and endoderm). They contain developmentally mature skin complete with hair follicles and sweat glands, sometimes luxuriant clumps of long hair and pockets of sebum, blood, fat, bone, nails, teeth, eyes, cartilage and thyroid tissue. MTs are usually benign, but undergo malignant transformation in <0.2% of cases, and reported incidence of 1–3% by Rim et al. (2006). Several malignancies may develop from any of the three germ-cell layers, such as adenocarcinoma, malignant thyroid struma, carcinoid tumors, melanomas and a variety of soft tissue sarcomas (Rim et al., 2006). The most common malignant evolution is squamous cell carcinoma (SCC) from ectoderm (Rim et al., 2006). Malignant melanoma arising within MCT is extremely rare, with an estimated incidence of <1%.

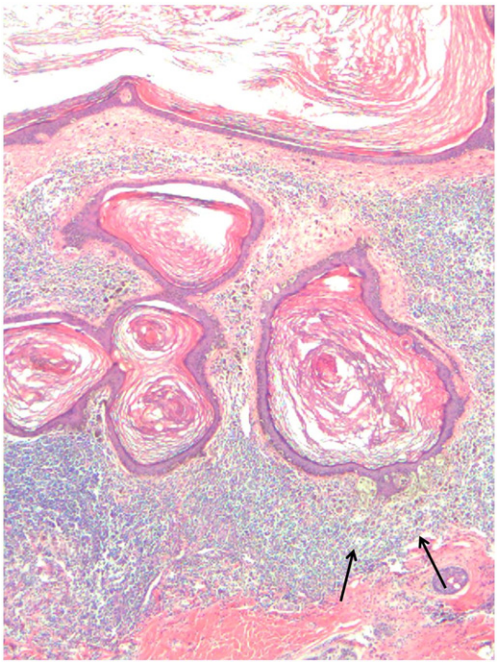

Fig. 1. Mature squamous epithelium in the ovarian teratoma contains numerous junctional nests of atypical melanocytes. Arrows mark an area shown at higher power (H&E stain, 4×).

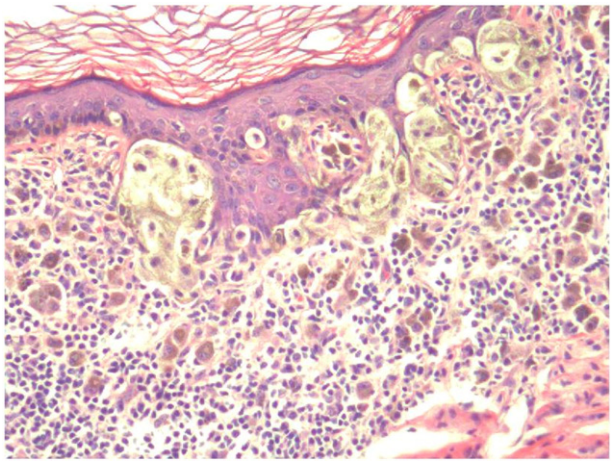

Fig. 2. Nests of melanoma in situ are present at the epithelial-stromal junction and invasive melanoma is present in the stroma accompanied by a prominent lymphocytic reaction (H&E, 20×).

We reviewed nine recent cases of malignant melanoma arising in a mature cystic teratoma reported during 2011–2015 (Table 1). The ages of the patients ranged 24–75 years, with an average of 51.1 years. Initial diagnosis was an ovarian mass in most cases. Four of nine patients had evidence of metastatic disease. The standard treatment for these patients was surgery, specifically unilateral salpingo-oophorectomy (USO) or total abdominal hysterectomy with bilateral salpingo-oophorectomy +/− staging procedure. Six of nine patients received adjuvant chemotherapy, while three patients additionally received immunotherapy. Like our case, two received no adjuvant therapy. Four patients had distant metastases, all of whom died of disease (DOD) at the time of publication. Out of five patients without metastasis, two were reported with NED and the follow-up for the other two was unknown (Table 1). From this, we can conclude distant metastasis plays a significant role in prognosis in a tumor of this type. Therefore, evaluation of the patient for metastases is of paramount importance, and either surgical staging or PET imaging should be considered in these patients. The possibility of a melanoma arising at another site, such as skin, central nervous system, eye and gastrointestinal tract with secondary involvement of the ovary tract, needs to be excluded.

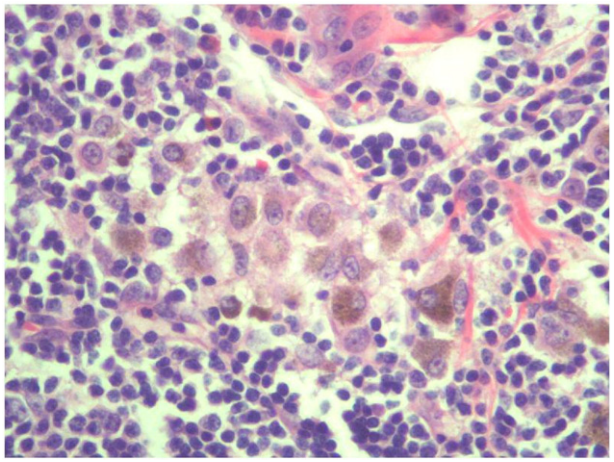

Fig. 3. Invasive melanoma (H&E, 40×).