



Ovarian teratoma with pituitary tissue: A case report

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ABSTRACT

Ovarian cysts and specifically ovarian teratomas are a common finding in young patients. These cysts display histological cell types from all three cell lines: endodermal, ectodermal and mesodermal origins. A 22-year-old woman who displayed classic signs of cortisol excess – excessive weight gain, difficulty losing weight and abdominal striae – was found to have a 10 cm mature teratoma cyst. This patient presented with ovarian torsion, a common complication of ovarian cysts, and was treated surgically. Pathology was significant for an ovarian teratoma with pituitary secreting cells, most significantly cells secreting adrenocorticotropic hormone (ACTH). © 2020 The Authors. Published by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

This is a unique case of an ovarian dermoid with mature pituitary tissue, which was positive for cells secreting growth hormone, prolactin and adrenocorticotropic hormone (ACTH), a rare pathological finding. Dermoid cysts, also known as benign cystic mature teratomas, are the most common ovarian tumor in women in the second and third decades of life. These cysts contain mature tissue of ectodermal, mesodermal, and endodermal origin; therefore, they are highly variable in form and histology [1]. Ovarian dermoid cysts most commonly exhibit several different types of tissue, such as bone, muscle and skin. It is rare to find more complex tissues within an ovarian dermoid such as brain, spinal cord, or eye tissue.

The pituitary glands' cell line precursor is the ectoderm, and it is able to secrete a variety of hormones that in turn may exert an effect on many other endocrine glands [2]. The pituitary gland is responsible for secreting endocrine hormones such as ACTH, follicle stimulating hormone (FSH), luteinizing hormone (LH), prolactin, thyroid stimulating hormone (TSH), Melanocyte-stimulating hormone (MSH) and growth hormone. Given that dermoid cysts can contain ectodermal tissue, it is possible that a dermoid cyst could contain pituitary tissue, but it is so rare that there is no current data available to give a rate of occurrence.

Often ovarian dermoid cysts are discovered when patients begin to exhibit pelvic or abdominal pain, bloating or constipation or present acutely with ovarian torsion [3]. Ovarian torsion occurs when the weight of an adnexal mass causes the ovary and fallopian tube to twist around the infundibulopelvic ligament, causing restriction of its

blood supply. Patients often present with acute onset of unremitting unilateral pelvic pain, nausea, vomiting and an adnexal mass [4].

2. Case Presentation

A 22-year-old woman presented to the emergency room with worsening lower left pelvic pain, nausea, vomiting and vaginal bleeding.

Her medical history was significant for a rapid weight gain the year prior, which prompted outpatient evaluation by an endocrinologist. She was reporting regular menses and sleep habits and denied hirsutism, acne or hair loss. Preliminary endocrine labs were reported as follows: HgbA1 4.8% [normal <5.7%], testosterone 29.9 [15–70 ng/dL], LH 8.1 [5–25 IU/L], FSH 4.1 [4.7–21.5 mIU/mL], TSH 2.41 [0.5–5.0 mIU/L], CMP normal, lipid panel normal. The patient attempted a trial of off-label metformin for weight loss with increasing exercise regimen; however, this was self-discontinued due to side-effects. Surgical and family history were noncontributory.

The patient attempted multiple therapies to lose weight, including injections of semaglutide, a GLP-1 receptor agonist, to aid an increase in insulin secretion. Endocrine visits were significant for abdominal striae; however, the patient was not tested for Cushing's syndrome as she did not display other symptoms, such as hirsutism, acne, buffalo hump, muscle wasting, heat intolerance or hypertension.

2.1. Diagnostic Assessment

On examination in the emergency room, the patient was found to be mildly tender over her left lower quadrant; however, no masses were appreciated secondary to body habitus. Transvaginal ultrasound and CT scan demonstrated a 10.6 cm multi-septated left adnexal mass,

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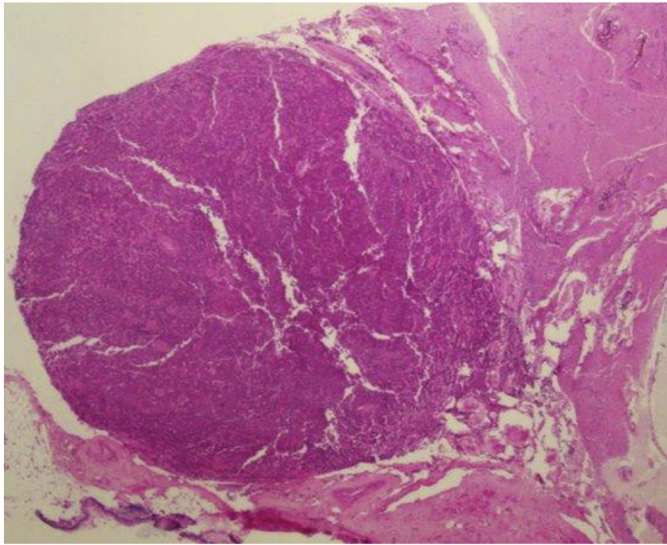


Fig. 1. Low-power view (4 \times), H&E: Showing well circumscribed nodule consisting of oncocytic cells.

containing fat, fluid, and calcifications, consistent with suspected dermoid cyst.

During the course of her emergency room visit, her pain improved and as ultrasound revealed arterial and venous blood flow to both ovaries, a decision was made for outpatient follow-up that week with a surgeon.

Shortly after her visit to the emergency room, the patient was seen in the outpatient clinic and scheduled for laparoscopic ovarian cystectomy. Upon laparoscopic entry, the left ovary was found to be torsed two times around its vascular pedicle with a large dermoid cyst. The dermoid was removed laparoscopically and residual ovarian parenchyma was repaired.

2.2. Follow-up and Outcome

The final pathology was a mature cystic teratoma (dermoid cyst) with a small focus of mature pituitary tissue. Per pathology review, identification of pituitary tissue was supported by a positive pancytokeratin, CAM 5.2, S100, and synaptophysin [seen in Figs. 1 and 2]. Stains for tropic hormones were positive for growth hormone, prolactin and ACTH [seen in Fig. 3].

The patient was seen one month after her surgery, when she was doing well and was noted to have modest weight loss since her operation. The patient was advised to have a repeat sonogram in 6 months, to check for a possible redevelopment of a teratoma. The patient has not been back to see her endocrinologist to date.

3. Discussion

Ovarian torsion affects women of all ages and is considered one of the more common gynecologic emergencies. This diagnosis encompasses either complete or partial rotation of the adnexa, which results in changes in the blood supply of the ovary, causing ischemia [3]. Risk factors include ovarian cysts, ovarian tumors, and fertility treatment [7]. Ovarian torsion has been reported to occur with masses ranging from 1 to 30 cm (mean 9.5 cm) in size, and often presents with acute lower abdominal or pelvic pain along with nausea and vomiting [5]. Treatment often includes laparoscopy to untwist the ovary or the adnexa and restore blood flow with a possible cystectomy to prevent torsion from recurring.

Mature ovarian teratomas are one of the most common benign ovarian neoplasms and include differentiated germ cell layers (ectoderm,

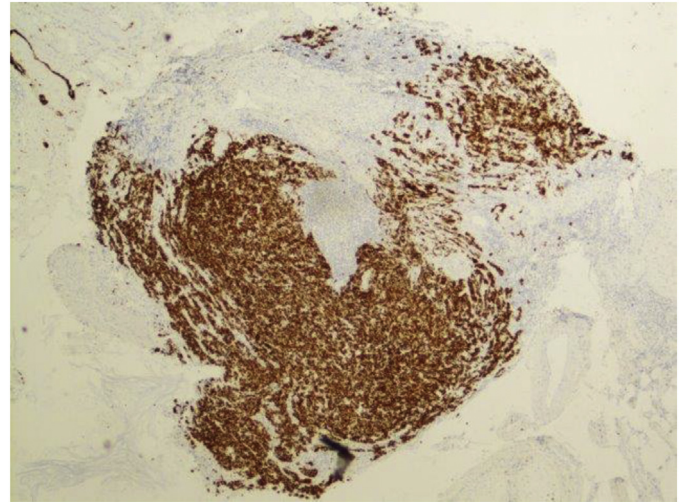


Fig. 2. Low-power view (4 \times): Positive staining for S-100 immunohistochemistry.

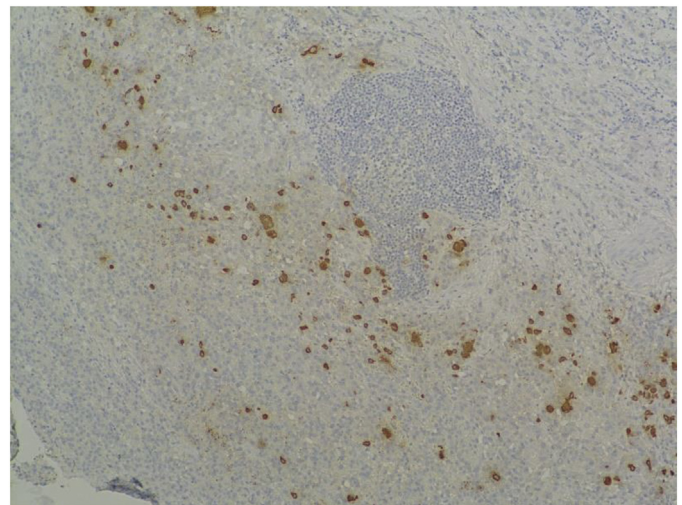


Fig. 3. Low-power view (100 \times) showing ACTH positive staining tumor cells (in brown). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

mesoderm, endoderm) and often are identified at the time of surgery after a patient presents with an ovarian torsion [6]. This is a case of ovarian torsion secondary to an ovarian mature teratoma that was later found to mature pituitary tissue with ACTH-secreting cells. ACTH normally is secreted by the pituitary gland, but has been found to be secreted from ectopic sources [8]. ACTH stimulates the adrenal gland to produce cortisol [2]. If ACTH is secreted in excess it can cause an increase in cortisol secretion, which can be associated with the symptoms this patient was experiencing, including acute weight gain and abdominal striae [2].

A limitation of this case report is that there was no laboratory confirmation of systemic ACTH or cortisol excess; rather, this patient's diagnosis was from clinical exam findings alone. When managing endocrine disorders, it is of value to remember that benign and malignant tumors can secrete ectopic hormones that can impact the entire hormonal axis. This case report adds value to clinical practice because this patient's symptoms were secondary to a very rare presentation of ectopic ACTH secretion.

Contributors

Sarah Werner drafted the article.

Cristina Zottola contributed to revision of the article.

Jordan Steinberg contributed the pathology slides.

Barry Pearson contributed the pathology review.

Adi Katz was the surgeon involved in the case and contributed to revision of the article.

Conflict of Interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

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Patient Consent

Written informed consent for publication of her details was obtained from the patient prior to the writing of this case report.

Provenance and Peer Review

This case report was peer reviewed.

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