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Ovarian steroid cell tumor in pregnancy-a rare occurrence. Report of a case and review of the literature.

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Background

Steroid cell tumors of the ovary are very rare, especially during pregnancy, and they must be distinguished from luteoma of pregnancy.

Case

An 18 year old female with an adnexal mass discovered incidentally during Caesarean section.

Workup revealed the tumor to be a steroid cell tumor, exceedingly rare in pregnancy.

Conclusions

Ovarian steroid cell tumors, which are malignant one-third of the time, are difficult to distinguish from luteoma of pregnancy.

Introduction

Steroid cell tumors of the ovary represent less than 0.1% of all ovarian tumors. Of the three subtypes, steroid cell tumors, not otherwise specified (NOS), are the most common, accounting for approximately 60% of all steroid cell tumors^[1]. Since most of these tumors are diagnosed early, there is limited information about their recurrence rate and response to chemotherapy^[2].

When steroid cell tumors-NOS are hormone-producing, the patient may present with virilization, hyperestrogenism, and Cushing's syndrome may occur. Most steroid cell tumors occur in women over 40, and the occurrence in pregnancy is extremely rare. Steroid cell tumors are difficult to distinguish from luteoma of pregnancy, which is a more well-known but still uncommon phenomenon of pregnancy^[3].

Case Report

The patient was an 18 year old G3P1011 female at 38 weeks gestational age, admitted for induction of labor for poorly controlled gestational diabetes. Past obstetric history was significant for a previous cesarean delivery due to arrest of dilation, and a pregnancy complicated by a fetus with omphalocele, which was terminated at 13 weeks. She reported regular menstrual periods prior to pregnancy, and had no other contributory history. There were no abnormal findings on pelvic exam. There was no facial acne, or hirsutism. The patient underwent induction of labor with pitocin and a transcervical foley balloon. Due to a category 2 tracing remote from delivery, she underwent repeat cesarean delivery.

Upon inspection of the left adnexa at the time of the cesarean delivery, a large cystic and solid appearing mass, measuring 12x14 cm, was noted extending from the left ovary. This was removed from its pedicle on the ovary. The other ovary was unremarkable in appearance. The male infant was normal in appearance with Apgar scores of 8/8.

Pathology

The specimen received was soft, but solid, and measured 12 x 10 x 7 cm. On cut surface, the tumor was noted to be bright yellow and lobulated (fig 1). Histologically the tumor was composed of sheets of eosinophilic cells(fig 2) with areas of vacuolization (fig 3), consistent with lipid. There were up to 2 mitoses per ten high power fields. The tumor stained for calretinin, inhibin, and smooth muscle actin, and was negative for cytokeratin 7 and desmin. The diagnosis of steroid cell tumor was rendered.

Discussion

Steroid cell tumors of the ovary are rare, especially during pregnancy, with only a few reported cases. Steroid cell tumors are classified into three subtypes: steroid cell tumor not otherwise specified (NOS), stromal luteoma and Leydig cell tumor. Steroid cell tumor NOS is the most common, accounting for more than half of these tumors. These tumors usually have a greater age of onset, average age of presentation being 43 years, with no association with pregnancy^[4].

About 25% of the tumors have no hormonal manifestations, with patients experiencing no or vague symptoms such as abdominal distension and pain^[5]. Our patient falls into this category

because she had an asymptomatic presentation with incidental discovery of the tumor during caesarean section.

Steroid cell tumors occurring in pregnancy can be difficult to distinguish from luteoma of pregnancy. Steroid cell tumors are usually unilateral and yellow, compared to luteomas of pregnancy which are most often bilateral and brown^[2]. Steroid cell tumors are often positive for fat stains corresponding to the vacuolated cytoplasm, as was seen in our case, which helps favor that diagnosis.

Hayes and Scully found that 25-43% of steroid cell tumors are found to be malignant, and histologic examination can provide clues to accurately predict malignant behavior^[4]. They found that the most accurate predictor of malignant behavior is more than 2 mitotic figures per 10 high-power fields. Other features associated with malignant behavior are grade 2-3 atypia, necrosis, hemorrhage, and a diameter of > 7 cm^[4]. Tumor diameter greater than 7 cm is associated with malignancy 78% of the time. Our patient's tumor was about 12 cm with 2 mitoses per 10 high-power fields, indicative of a need for close follow-up.

There are no well-established protocols for treating this rare tumor. The primary mode of management is surgical excision, although chemotherapy has been utilized in some cases. The management of asymptomatic adnexal masses in pregnancy continues to be challenging as management guidelines are mainly based on case-control or observational studies^[6].

A review of the literature revealed two other cases of steroid cell tumors which were associated with pregnancy^[3,5]. The first case was a 20 year old female who had a full-term pregnancy 9 months after surgical intervention for steroid cell tumor NOS^[5]. The second case was a 24 year old female who was found to have an 8 cm solid, homogenous mass excised at delivery of a male fetus at 39 weeks by caesarean section^[3]. Both cases presented with virulization of the mother.

In conclusion, we describe a case of steroid cell tumor in pregnancy detected incidentally during caesarean section. The occurrence of this lesion in pregnancy is exceedingly rare, and the lesion must be distinguished from luteoma of pregnancy, which is a hyperplastic rather than neoplastic condition.

References:

1. Hasegawa K, Minami Y, Inuzuka H, et al. Female pseudohermaphroditism associated with maternal steroid cell tumor, not otherwise specified of the ovary: a case report and literature review. *Clin Exp Obstet Gynecol.* 2013;40:591-595.
2. Burandt E, Young RH. Pregnancy luteoma: a study of 20 cases on the occasion of the 50th anniversary of its description by Dr. William H. Sternberg, with an emphasis on the common presence of follicle-like spaces and their diagnostic implications. *Am J Surg Pathol* 2014; 38: 239-244.
3. Oz, M., Ozgu, E., Turker, M., Erkaya, S., & Gungor, T. Steroid cell tumor of the ovary in a pregnant woman whose androgenic symptoms were masked by pregnancy. *Arch Gynecol Obstet* 2014; 290: 131-134.
4. Hayes MC, Scully RE. Ovarian steroid cell tumors (not otherwise specified). A clinicopathological analysis of 63 cases. *Am J Surg Pathol.* 1987;11:835–845
5. Sielert L, Liu C, Nagarathinam R, Craig LB. Androgen-producing steroid cell ovarian tumor in a young woman and subsequent spontaneous pregnancy. *J Assist Reprod Genet.* 2013; 30:1157-1160.

6. Goh W, Bohrer J, Zalud I. Management of the adnexal mass in pregnancy. *Curr Op Obstet Gynecol.* 2014; 26: 49-53.

7. Haroon S, Zia A, Idrees R, Memon A, Fatima S, Kayani N. Clinicopathological spectrum of ovarian sex cord-stromal tumors; 20 years' retrospective study in a developing country. *Journal of Ovarian Research.* 2013;6:87. doi:10.1186/1757-2215-6-87.

8. Jiang W, Tao X, Fang F, Zhang S, Xu C. Benign and malignant ovarian steroid cell tumors, not otherwise specified: case studies, comparison, and review of the literature. *Journal of Ovarian Research.* 2013;6:53. doi:10.1186/1757-2215-6-53.

