

Pregnancy Luteoma: A Case Report

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1. Abstract

Pregnancy luteoma is a non-neoplastic lesion of the ovary related to hormonal effects of pregnancy that is usually discovered incidentally at the time of a caesarean section or during postpartum tubal ligation. We herein report a case of a 33-year-old full-term pregnant female who presented with abdominal pain and a right-sided ovarian mass.

2. Introduction

Luteoma of pregnancy is a rare ovarian lesion, first described by Sternberg in 1963 [1] and thought to arise from excessive response of ovarian stromal cells to pregnancy hormones, especially beta-human chorionic gonadotropin (β -hCG) [2]. Pregnancy luteoma represents a diagnostic and therapeutic challenge in that it can mimic a malignant ovarian neoplasm [3]. As pregnancy luteoma usually regresses spontaneously postpartum [1] a high clinical suspicion is mandatory for appropriate therapeutic management.

3. Case Report

The authors describe a case of a pregnancy luteoma in a 33-year-old full-term pregnant woman. A 31-year-old woman (G1P0) was admitted to the Obstetrics and Gynecology Department in the University Hospital of Patras with abdominal pain. She had a medical history of polycystic ovarian syndrome (PCOS), diabetes mellitus type I, high blood pressure and an increased body mass index (BMI=37,5). She didn't have an antenatal care on a regular basis. An abdominal ultrasound revealed a right-sided ovarian mass.

No previous clinical information regarding this adnexal mass was mentioned, so that a caesarean section was performed. During the caesarean section a palpable mass of the right ovary was found; an ovarian mass excision was carried out and the specimen was submitted to histopathological analysis. A male newborn was delivered with a birth weight of 2960 gr, which was not affected by his sex.

Macroscopic examination showed a well circumscribed 3.4cmx2cmx2cm mass that on cut section was solid, soft, fleshy and grey to grey brown. Hematoxylin and eosin stained paraffin sections revealed a multinodular solid mass composed of round to polygonal cells arranged in sheets, islands and cords with round to oval vesicular nucleus with variably prominent nucleoli and abundant eosinophilic granular cytoplasm. Nodules were separated by thin fibrous septa and there were numerous capillaries. Up to 7 mitotic figures per 10 HPF were found. Rare lipid containing cells were identified while nuclear atypia was mild and there was no necrosis, nuclear grooves or Reinke crystals. On immunohistochemistry cells expressed calretinin, inhibin, vimentin, Melan A while other markers (HMB45, S-100 etc) were negative. Reticulin stain showed fibers surrounding groups of cells. Based on the above histopathologic findings and the clinical history of pregnancy other sex-cord stromal tumors of the ovary, mainly Leydig cell tumor, steroid cell tumor NOS (especially the lipid poor variant) and luteinized granulosa cell tumor were excluded and a final diagnosis of pregnancy luteoma was made.

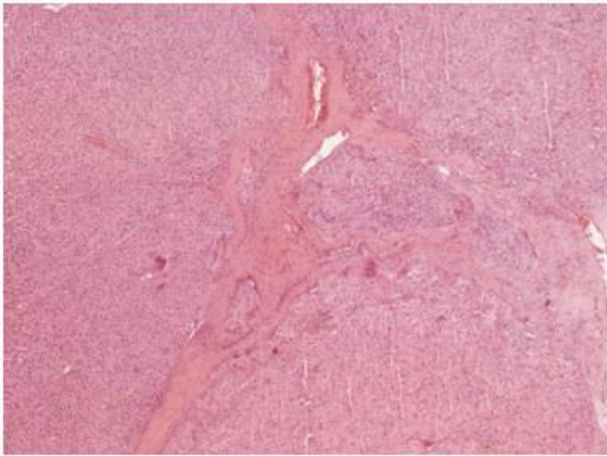


Figure 1A: Light microscopy shows a solid mass of nodules separated by fibrous septa. (hematoxylin and eosin stain, x40).

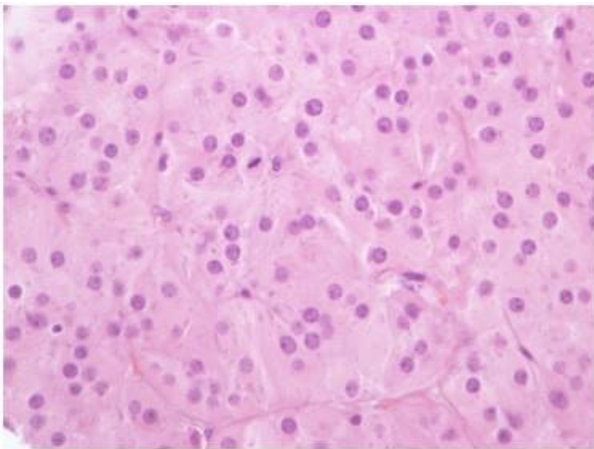


Figure 1B: The lesion is composed of polygonal cells with variably prominent nucleoli and abundant eosinophilic granular cytoplasm (hematoxylin and eosin stain, x400).

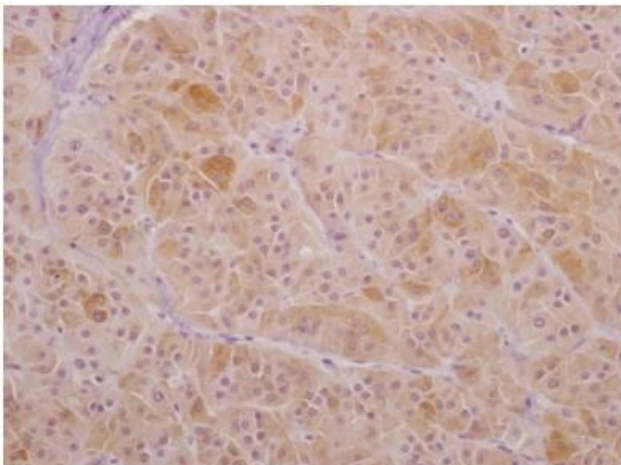


Figure 1C: On immunohistochemistry cells show positive staining for inhibin (hematoxylin and eosin stain, x200).

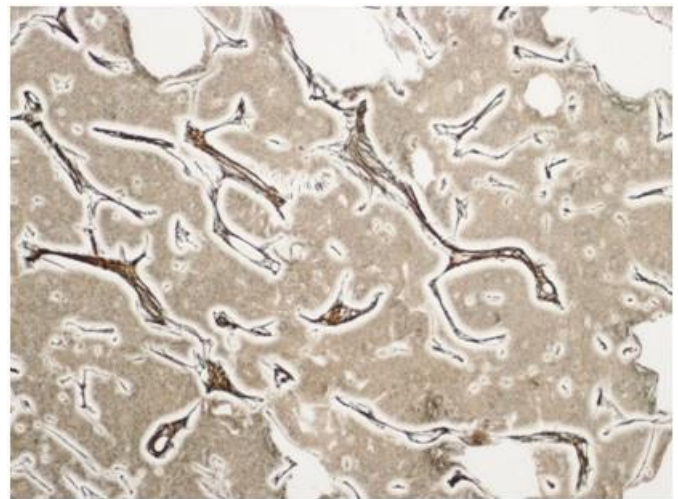


Figure 1D: Reticulin staining (x200).

4. Discussion

Pregnancy luteoma is a relatively rare lesion as fewer than 200 cases have been reported in the literature [2,4]. Most cases are incidental findings during caesarean section or tubal ligation, but occasionally ultrasound antenatal diagnoses have been made [2]. Pregnancy luteomas are thought to be caused by hyperplasia of luteinized stromal cells secondary to stimulation by beta-human chorionic gonadotropin (β -hCG) [5]. In women with PCOS, stromal cell hyperplasia may antedate pregnancy [6]. However, β -hCG appears unlikely to be the only etiological factor, because the lesions are not reported in gestational trophoblastic disease or early pregnancy, when β -hCG levels are highest [2,7]. Clinically, luteomas are often silent and only discovered incidentally during peripartum surgery. In 25% of cases, luteomas are hormonally active, secreting androgens, which can result in maternal hirsutism and fetal virilization [8]. Virilization of the female fetus occurs in half of the patients with maternal hirsutism, which results in clitoral enlargement and ambiguous genitalia. Fetal sensitivity to maternal serum androgen depends on both the age at which exposure occurs and the ability of the placenta to aromatize androgens into estrogens. Male fetuses are not affected by this condition [9-11]. Luteomas represent a diagnostic and therapeutic challenge because they can mimic a malignant ovarian tumor. The differential diagnosis for pregnancy luteomas includes granulosa cell tumors, thecomas, Sertoli-Leydig cell tumors, pure Leydig (hilar) cell tumors, stromal hyperthecosis, stromal luteomas, and hyperreaction luteinalis. Because of the solid nature of the mass, it is impossible to differentiate luteomas from other solid ovarian neoplasms such as luteinized thecoma, granulosa cell tumor, or Leydig cell tumor based on imaging characteristics alone [12]. Pregnancy luteomas vary in size, ranging from microscopic to over 20cm in diameter [13,14]. On gross examination, cut surfaces of luteomas are solid, soft, tan or flesh colored, with hemorrhagic foci. Microscopically they contain large groups of eosinophilic cells surrounded by numerous blood vessels. Sometimes a cord-like pattern or follicular

arrangement with colloid nest is noted. Cells are intermediate in size between granulosa lutein and theca lutein cells and contain vacuolated cytoplasm. Intracellular lipids are rarely seen [13]. On electron microscopy, these cells contain abundant smooth endoplasmic reticulum, dispersed Golgi apparatus, and tubular cristae in mitochondria, similar to other steroid producing cells [15]. On histologic grounds other sex cord-stromal tumors of the ovary, including mainly Leydig cell tumor, steroid cell tumor NOS (especially the lipid poor variant) and luteinized granulosa cell tumor, enter the differential diagnosis so that clinical history is very important in order to reach correct diagnosis of pregnancy luteoma. The management of a suspected pregnancy luteoma depends on the clinical situation and the woman's desires. If the mass is found in the second trimester with size greater than 5cm, it is reasonable to either observe or perform surgical exploration to eliminate risk of torsion, obstruction and rupture [16]. If conservative management is opted, patients should be evaluated postpartum, because pregnancy luteomas usually resolve 2-3 weeks postpartum.

5. Conclusion

Pregnancy luteoma represents a benign pregnancy-related condition that generally resolves spontaneously after delivery. In most cases, it is asymptomatic and is accidentally detected during caesarean section. High clinical suspicion of pregnancy related lesions/tumors and intraoperative examination of ovaries, the fallopian tubes and the appendix for abnormalities is recommended.

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