



Luteoma of Pregnancy with Uterine Leiomyoma as a Diagnostic Challenge and Mimicker of Ovarian Malignancy: A Rare Case From Rural India

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Abstract

Background: Luteoma of pregnancy is a rare, non-neoplastic lesion of the ovary which mimics ovarian tumor. It develops hirsutism or virilization during pregnancy and regresses in postpartum phase spontaneously. A few number of cases are described in literature. The synchronization of ovarian luteoma with uterine leiomyomas is a rare condition; to the best of my knowledge, the association of both conditions is not reported till date.

Case Presentation: A 35 year old multiparous woman presented with mass and pain in abdomen came to the gynecology department of Uttar Pradesh University of Medical Sciences in India. On examination, a hard mass in uterus was diagnosed. Ultrasonography revealed a highly vascular uterine adnexal mass possibly arising from ovary with malignant features. Hysterectomy with bilateral salpingo-oophorectomy was done and this specimen with separate mass was sent for histopathology. Right sided ovary was measured 3×3 cm, well demarcated with solid brown areas. CA -125 (cancer antigen -125) level was raised. A separate tumor was measured 15×12×8 cm and it was smooth while cut surface was grey/white with whirling and hemorrhagic foci.

Results: The section from ovary revealed luteoma of pregnancy. Endometrium microscopy confirmed pregnancy. Section from another mass/tumor showed leiomyoma with degenerations.

Conclusion: Pregnancy luteoma is a pregnancy induced lesion which mimics malignancy so, it is clinically misinterpreted and over diagnosed. Histopathological investigation is mandatory to diagnose and prevent unnecessary surgeries. The synchronous ovarian luteoma with uterine leiomyoma is a rare condition which represents unusual response to altered hormonal effect in pregnancy; therefore, more studies should be done to understand its pathogenesis.

Keywords: Histopathology, Leiomyoma, Luteoma of pregnancy.

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Introduction

Luteoma of pregnancy is a rare non neoplastic hormone dependent lesion of the ovary that occurs in pregnancy and regresses after delivery (1). It is usually discovered and diagnosed at the time of cesarean section. It was first described by Sternberg and Barclay in 1966 (2). Hyper-secretion of androgens happens in some cases causing virilization in mother and fetus. On clinical

examination and ultrasound, it is usually present as heterogenous adnexal mass and often misdiagnosed as malignancy (3). Leiomyoma of the uterus is a benign neoplasm of reproductive age group (4). The coincidence of ovarian luteoma with uterine leiomyoma is a rare finding which demonstrates the impact of hormones as a predisposing factor. In this study, a 35 year old multipa-

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rous woman was reported with clinical diagnosis of ovarian malignancy but on histopathological examination, luteoma of ovary with leiomyoma of the uterus was detected. To the best of my knowledge, the association of luteoma of pregnancy with leiomyoma of the uterus has not been reported in literature till date.

Case Presentation

A 35 year old multiparous women (Gravida 5, para 5, and 5 live births) presented with mass and pain in abdomen to Outdoor Patients Department of Uttar Pradesh University of Medical Sciences of India in 2016. The patient was lean and thin. There was no other significant medical history and no information regarding her current pregnancy. On clinical examination, a hard mass in uterus was diagnosed. The patient did not have PCOS, and there was also no history of previous luteoma. Ultrasonography revealed a highly vascular adnexal mass possibly arising from ovary. Her CA-125 level was raised. Hysterectomy with bilateral salpingo-oophorectomy results together with separate mass in 10% formal saline were sent to histopathology section of pathology department and simultaneously endometrial biopsy was sent for histopathology examination. The frozen section technique was not done as it was not possible at that time. On gross examination, uterus with mass was measured $20 \times 13 \times 10$ cm whereas tumor was measured $15 \times 12 \times 8$ cm. On sectioning, growth was well defined and cut surface was grey/white with whirling and hemorrhagic foci. The bilateral ovaries were solid and homogenous with grey/white cut surface. Right sided ovary was 3×3 cm, well demarcated with solid brown areas. Multiple sections were taken at 2-4 micron and stained with H&E and PAS. Section from endometrium showed chorionic villi with cytotrophoblast and syncytiotrophoblast confirming retained products of conception (Figure 1). The section from ovary showed groups of luteinized polyhedral cells with pink granular cytoplasm, round to oval prominent nuclei with mild atypia and absence of typical Reinke's crystalloids (Figure 2). PAS stain was also applied (Figure 3). Section from growth showed numerous smooth muscle fibers running at various angles in between hemorrhagic foci. Hyaline, hydropic, and fatty degeneration (Figure 4) was also detected. Based on these findings, the case was diagnosed as luteoma of pregnancy with uterine leiomyomas.

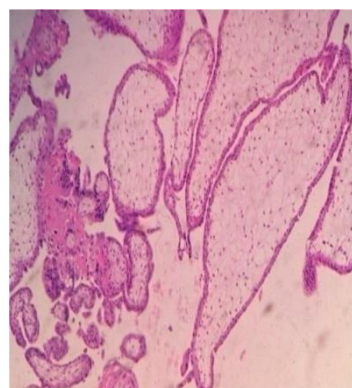


Figure 1. H&E stained section of retained products of conception

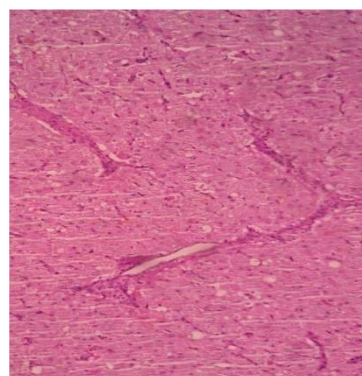


Figure 2. H&E stained section of luteoma

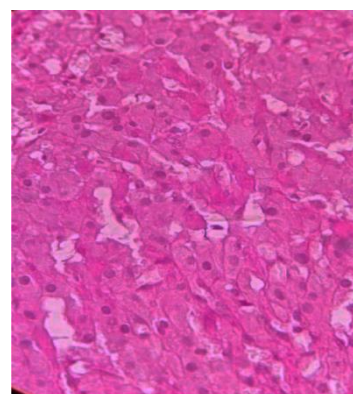


Figure 3. PAS stained section of luteoma

Discussion

Pregnant women rarely have adnexal masses and the prevalence of the problem is between 1 to 2%. They are usually diagnosed at routine antenatal check-ups. The common adnexal masses are simple cysts, hemorrhagic cysts, leiomyomas and hyperstimulated ovaries in patients who had gone through assisted fertility (5). Hyperreactio lutein

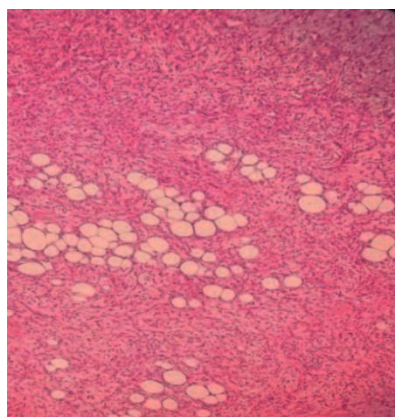


Figure 4. H&E stained section of uterine leiomyoma

alis and luteoma are few adnexal masses associated to pregnancy. Luteoma of pregnancy is a rare non neoplastic lesion of ovary which occurs during pregnancy and regresses spontaneously after delivery. It is usually asymptomatic and found as incidental finding on caesarean section or postpartum tubal ligation. It is thought to arise from excess pregnancy secretion but not found in pathologies like trophoblastic disease and hyperreactio luteinalis suggesting other unknown contributing factors in its development, while there are few predisposing factors such as multiple pregnancies, advanced maternal age, polycystic ovary syndrome, and previous history of luteoma (6, 7). It occurs during the third and fourth decades of life and about 80% are associated with multiparous women and may cause maternal virilization in 25-30% of women; they might also be risky for 50% of female fetuses (8). Pregnancy luteomas are variable in size, ranging from microscopic to over 20 cm in diameter. They are often bilateral in one third of cases and may cause mass effects on adjacent organs like ureter, or lead to obstructive uropathy and ovarian scarring. In the present case, the woman was 35 years old having five live children. Luteoma of the ovary was unilateral with 3 cm in largest diameter. On ultrasonography, they often appear as heterogeneous solid masses, predominantly hypoechoic with thick wall. Most of the time, luteoma of ovary is highly vascular and may mimic ovarian malignancy. There are also rare reports of pregnancy luteomas presenting like malignant tumors with raised CA-125 level (9). In the present case, CA-125 level was also raised, and case was clinically and radiologically suspicious of ovarian malignancy. Regarding the gross appearance of luteoma, cut surface was solid, tan

or flesh colored with hemorrhagic foci (3), whereas microscopy revealed sharply circumscribed nodules of polygonal cells arranged in sheets, cords or small follicles having colloid like substance. The cytoplasm is eosinophilic and finely granular and nuclei may show slight pleiomorphism (1, 10). These findings were consistent in the present study. Various types of degenerations are common in large leiomyomas (11) and occur because of decreased vascularity (12). Different types of degeneration were seen in the current case in leiomyoma along with leutoma.

Conclusion

Pregnancy luteoma is a tumor like lesion seen during pregnancy and mimics malignancy. Histopathological investigation is mandatory to diagnose the problem and prevent unnecessary surgeries. The synchronous ovarian luteoma with uterine leiomyoma is a rare condition; therefore, more studies should be done to understand the pathogenesis.

Conflict of Interest

Nil.

References

1. Limaïem F, Bouraoui S, Bouslama S, Lahmar A, Mzabi S. A puzzling ovarian tumor pregnancy luteoma with diffuse endometriosis. *Pathologica*. 2016;108(3):144-7.
2. Spitzer RF, Wherrett D, Chitayat D, Colgan T, Dodge JE, Salle JL et al. Maternal luteoma of pregnancy presenting with virilization of the female infant. *J Obstet Gynaecol Can*. 2007;29(10):835-840.
3. Chiang G, Levine D. Imaging of adnexal masses in pregnancy. *J Ultrasound Med*. 2004;23(6):805-19.
4. Mohan H. Textbook of pathology. 6th ed. New Delhi: Jaypee brothers medical publishers; 2010. Chapter 24, The female genital tract; p. 721-53.
5. Nelson MJ, Cavalieri R, Graham D, Sanders RC. Cyst in pregnancy discovered by sonography. *J Clin Ultrasound*. 1986;14(7):509-12.
6. Kao HW, Wu CJ, Chung KT, Wang SR, Chen CY. MR imaging of pregnancy luteoma: a case report and correlation with the clinical features. *Korean J Radiol*. 2005;6(1):44-6.
7. Phelan N, Conway GS. Management of ovarian disease in pregnancy. *Best Pract Res Clin Endocrinol Metab*. 2011;25(6):985-92.
8. Zander J, Mickan H, Holzmann K, Lohe KJ. Androluteoma syndrome of pregnancy. *Am J Obstet Gynecol*. 1978;130(2):170-7.

9. Rodriguez M, Harrison TA, Nowacki MR. Leutoma of pregnancy presenting with massive ascitis and markedly elevated CA125. *Obstet Gynecol.* 1999;94 (5 Pt 2):854.
10. Verma V, Paul S, Chahal KS, Singh J. Pregnancy leutoma: a rare case report. *Int J Appl Basic Med Res.* 2016;6(4):282-3.
11. Rosai J. Text book surgical pathology. 9th ed. New Delhi: Elsevier; 2004. chapter 19, Leomyoma; p. 1603-8.
12. Dayal S, Kumar A, Verma A. Clinico pathologic correlation of Leiomyoma with clinical findings and secondary changes in a rural population of North India. *Am J Clin Pathol.* 2014;141(2):275-9.