International Journal of Reproduction, Contraception, Obstetrics and Gynecology
Chinnathambi PS et al. Int J Reprod Contracept Obstet Gynecol. 2022 Oct;11(10):2850-2852
www.ijrcog.org
pISSN 2320-1770 | eISSN 2320-1789

DOI: https://dx.doi.org/10.18203/2320-1770.ijrcog20222488

Case Report

Tubal lymphangioma associated with ectopic pregnancy: a rare case report

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Received: 10 August 2022
Accepted: 03 September 2022

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ABSTRACT

Cavernous lymphangioma is a rare benign lymphatic neoplasm which occurs in occur in oral cavity, limbs, and abdomen. Presentation of cavernous lymphangioma of the fallopian tube is extremely rare. Cavernous lymphangioma presenting in a case of ectopic pregnancy is discussed below.

Keywords: Lymphangioma, Fallopian tube, Adnexal mass

INTRODUCTION

Tumors of lymphatic vessels comprise 4% of all vascular tumors.[1] Cavernous lymphangioma is a rare benign lymphatic neoplasm which may occur in oral cavity, limbs and abdomen.[1,2] Occurrence of cavernous lymphangioma in fallopian tube is extremely rare, which may be noted incidentally during laparotomy for other reasons. One such case of cavernous lymphangioma presenting with signs and symptoms of tubal ectopic pregnancy is discussed below.

CASE REPORT

A 23-year-old woman presented to the obstetrics clinic with complaint of pain in lower abdomen for 2 days. She had a history of one month amenorrhea and tested positive for b-HCG levels in urine, a day prior to the onset of pain. On general examination, she was afebrile with stable vitals. Abdomen was soft and tenderness was noted in the suprapubic region. Ultrasonography of the pelvis showed a heterogenous hypoechoic mass in left adnexa, measuring 4.6×3.5 cm, and minimal fluid in the pelvis thereby suggesting a possibility of aborted tubal ectopic pregnancy. Emergency laparotomy was carried out. Left tube was found to be edematous and congested with bleeding from the fimbrial end. There were blood clots in the pouch of Douglas amounting to 2×2 cm. Left ovary was normal. Right tube and ovary were normal. Left salpingectomy was done, and the tube was received in the pathology laboratory.

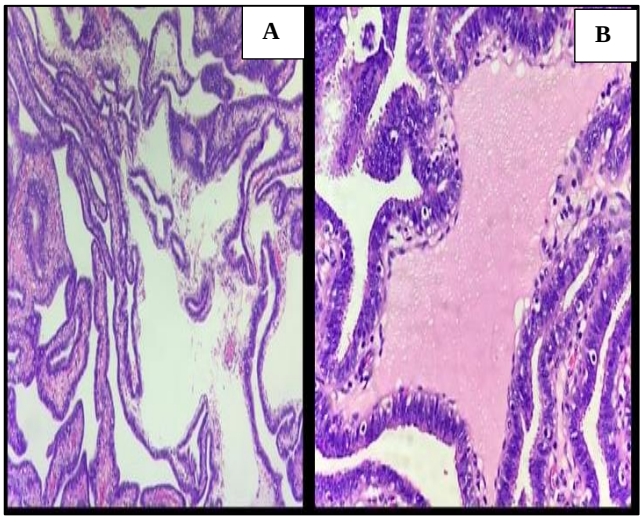

Figure 1 (A and B): Microphotographs showing fallopian tubal plicae with dilated lymphatic channels, bearing pink fluid at places (stain: H and E, 4X and 10X).

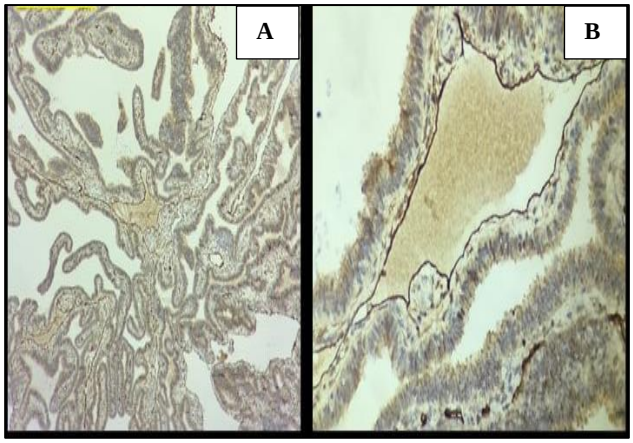

Figure 2 (A and B): Microphotographs showing fallopian tubal plicae with dilated lymphatic channels, endothelial lining of which is highlighted by D2-40 antibody (stain: IHC, 4X and 10X).

Grossly the tube was 7.2 cm long, enlarged with smooth external surface. Cut section showed tiny tubal papillary projections in the lumen. The entire specimen was embedded. Microscopy showed tubal plicae lined by ciliated columnar cells while the underlying stroma showed dilated lymphatic channels filled with proteinaceous pink fluid material at places (Figure 1 A and B). No evidence of chorionic villi or trophoblastic tissue noticed in the submitted sections. With a suspicion of cavernous lymphangioma, immunohistochemistry analysis was carried out with D2-40 antibody which showed a high intensity of staining in the endothelial cells of the lymphatic channels (Figure 2 A and B). A final diagnosis of cavernous lymphangioma of the left fallopian tube was rendered, with a follow up advice of b-HCG evaluation along with ultrasound scan of uterus and the other fallopian tube. HCG levels returned to normal range, with normal scan findings in uterus and the right adnexa.

DISCUSSION

Few queries had to be answered in the present case. Was there a conception? If so, where had the conception occurred and why the tube had not shown any evidence of decidualization changes or trophoblastic villi? These questions came into our discussion because there was a positive UPT with B-HCG levels of 3000 mIU/ml prior to surgery which returned to 3.2 mIU/L post-surgery. So, conception had occurred, however, if it had occurred in the left fallopian tube, there should have been evidence for the same, which was not found in this case. Moreover, the right tube was normal. The free fluid in the pouch of Douglas with some blood clots in it and the bleeding from the fimbrial end suggested a very early left tubal pregnancy. This pregnancy may have been missed or may have not progressed possibly due to the cavernous lymphangioma in the fallopian tube. This was later confirmed by return of the b-HCG levels to normal post-surgery, consolidated by the normal uterine and tubal scan findings by ultrasonogram.

Lymphangiomas are benign soft tissue tumors usually occurring in children less than 2 years of life.[1] It is closely related to cystic hygroma, which presents as a large lesion in the head and neck of babies. Lymphangiomas in oral cavity, limbs and abdomen has been documented.[1,2] But the occurrence of lymphangioma in fallopian tube is extremely rare.[3] Occurrence of tubal lymphangioma may hinder the progress of early gestation. Doddareddy et al had reported a case of tubal lymphangioma in a 32-year-old woman in which radiological findings suggested an endometriotic cyst or a leaking ectopic.[4] They had ruled out ectopic pregnancy by investigating b-HCG levels which was normal (<5 mIU/ml). Akbulut et al had reported a case of cystic lymphangioma of the fallopian tube, in a 69-year-old post-menopausal woman. According to them, all lymphangiomas are benign in nature but they can compress the vital structures.[5] Nigam et al has reported a case of lymphangioma of fallopian tube, in which the patient presented with fever, foul smelling vaginal discharge and bleeding. The clinical diagnosis in their case was septic abortion with adhesions which turned out to be tubal lymphangioma on histopathology.[6]

CONCLUSION

The uniqueness of the present case is the mystery associated with a lymphangioma that was incidentally diagnosed on a specimen clinically suspected to be a tubal pregnancy. Extensive search for tubal pregnancy yielded no evidence of tubal gestation despite positive biochemical and sonological findings leading to the assumption that a fimbrial ectopic was shed into the abdominal cavity or a uterine pregnancy may have been missed. The role of pathologist as a detective is substantiated in this very interesting case. Further it adds to the speculation that the progression of ectopic pregnancy was halted by the presence of lymphangioma. Hence awareness of this entity and a careful evaluation with a keen eye of speculation is required in cases of enlarged adnexal mass.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

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Cite this article as: Chinnathambi PS, Afsar N. Tubal lymphangioma associated with ectopic pregnancy-a rare case report. Int J Reprod Contracept Obstet Gynecol 2022;11:2850-2.