

Hilus cells in tuba uterina; an uncommon localisation and coincidence with endometrial carcinoma: A case report and review of the literature

Tuba uterina'da hilus hücreleri; sıradışı bir yerleşim ve endometriyal karsinom ile birliktelik: Olgu sunumu ve literatürün gözden geçirilmesi

Alp USUBÜTÜN

Hacettepe Üniversitesi Tıp Fakültesi Patoloji Anabilim Dalı, ANKARA

ABSTRACT

We present a patient having an endometrial carcinoma with an incidental finding of hilus cells in the tuba uterina. To our knowledge this is the first report of an incidental finding of hilus cells in the fallopian tube coincident with an endometrial carcinoma. No gross abnormality was seen in the salpinges, but microscopic examination showed scattered aggregates of hilus cells beneath a normal tubal epithelium. Both ovaries showed prominent hilus cells in the hilus also. Immunohistochemical findings were as follows; inhibin, calretinin, Melan A, vimentin were positive and EMA, CD99 were negative. Both microscopic and immunohistochemical features were identical to hilus cells of the ovary. So immunohistochemistry may be a useful tool to differentiate the extra-ovarian hilus cells from their mimics.

Key words: Hilus cell, endometrial carcinoma, immunohistochemistry, tuba uterina

ÖZET

Endometriyum adenokarsinomu olan bir hastada rastlantısal olarak tuba uterinasında hilus hücreleri saptanan bir olgu sunulmaktadır. Bildiğimiz kadarıyla olgumuz literatürde tuba uterinasında hilus hücreleri saptanan endometriyum karsinomlu ilk olgu sunumudur. Makroskopik olarak hiç bulgu saptanmamakla beraber, mikroskopik incelemede normal tuba epiteli altında kümeler halinde yayılmış hilus hücreleri görüldü. Ayrıca her iki over hilusunda da yaygın hilus hücreleri vardı. İmmünohistokimyasal olarak inhibin, kalretinin, Melan A, vimentin pozitif ve EMA, CD99 ise negatif olarak saptandı. Hem mikroskopik hem de immünohistokimyasal bulgular, overin hilus hücreleri ile özdeşti. İmmünohistokimya over dışı hilus hücrelerini benzerlerinden ayırmak için yararlı bir araç olarak kullanılabilir.

Anahtar sözcükler: Hilus hücresi, endometriyal karsinom, immünohistokimya, tuba

INTRODUCTION

Hilus cells are ovarian counterpart of testicular Leydig cells, with the exception of female chromatin pattern. These cells are round to oval and may contain crystals of Reinke (1). Hilus cells are present during fetal life, not identi-

fied in childhood but reappear at puberty and are demonstrable in all postmenopausal women at the hilar region of the ovary (2). Few cases have reported the existence of hilus cells outside the ovarian hilus, especially in the fallopian tube (3,4,5). None of the cases reported in the literature were associated with a neoplastic process. We present a patient having an endometrial carcinoma with an incidental finding of hilus cells in the tuba uterina.

Corresponding Author: Dr. Alp Usubütün, Hacettepe Üniversitesi Tıp Fakültesi Patoloji AD Sıhhiye 06100, Ankara

CASE REPORT

A sixty-six-year old woman referred to our hospital because of her endometrial carcinoma. She had an abdominal pain and vaginal bleeding for 2 months and the endometrial biopsy that was done in another center had reported grade 1, endometrioid type endometrial carcinoma. She had diabetes mellitus and otherwise was a healthy woman. Serum CEA, CA15.3, CA 19.9 and CA125 levels were normal. Total hysterectomy, bilateral salpingo-oophorectomy, pelvic and paraaortic lymphadenectomy and omentectomy were done.

On macroscopic examination, the uterus was 145 gr in weight and 8.5x4.5x4 cm in size.

Cut section of the uterus showed a tumor with both polypoid and infiltrative features. The depth of tumor infiltration measured 1.5 cm in

the myometrium and the intact myometrium was 0.5 cm in this area. No other gross abnormality was detected including the salpinges, ovaries, omentum and lymph nodes. Both salpinges were sampled completely by cutting them into two along the long axis.

No omental and lymph node involvement was detected. The interesting finding detected on left salpinx was aggregates of hilus cells on the fimbrial part. These aggregates were scattered beneath a normal tubal epithelium (Figure 1A). The cells forming these aggregates were large with oval to round eosinophilic cytoplasm and vesicular nuclei containing prominent nucleoli (Figure 1B). No prominent atypia or mitosis was found. These hilus cell aggregates were rich in vascular structures. Both ovaries also showed prominent hilus cells in their hili as seen in some postmenopausal cases. (Figure 1C).

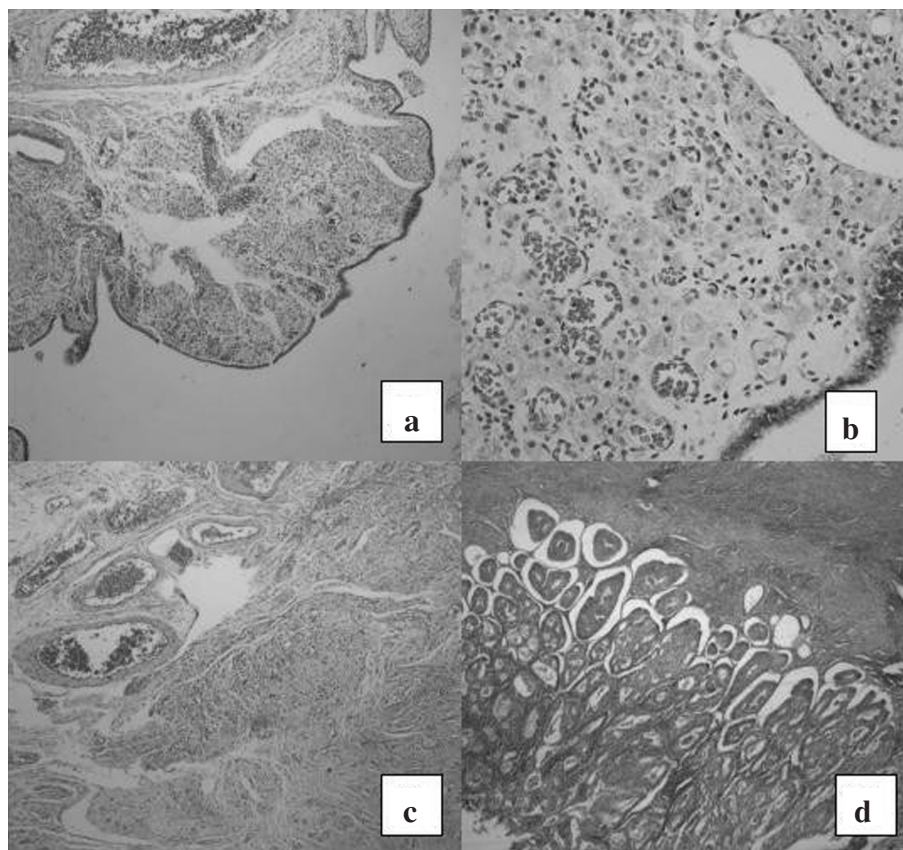


Figure 1. a) Aggregates of hilus cells were scattered beneath the tubal epithelium (x40). b) The cells have large oval or round eosinophilic cytoplasm and vesicular nuclei containing prominent nucleoli (x400). c) Both ovaries showed prominent hilus cells in the hili (x200). d) A well differentiated endometrioid carcinoma of the endometrium (x40).

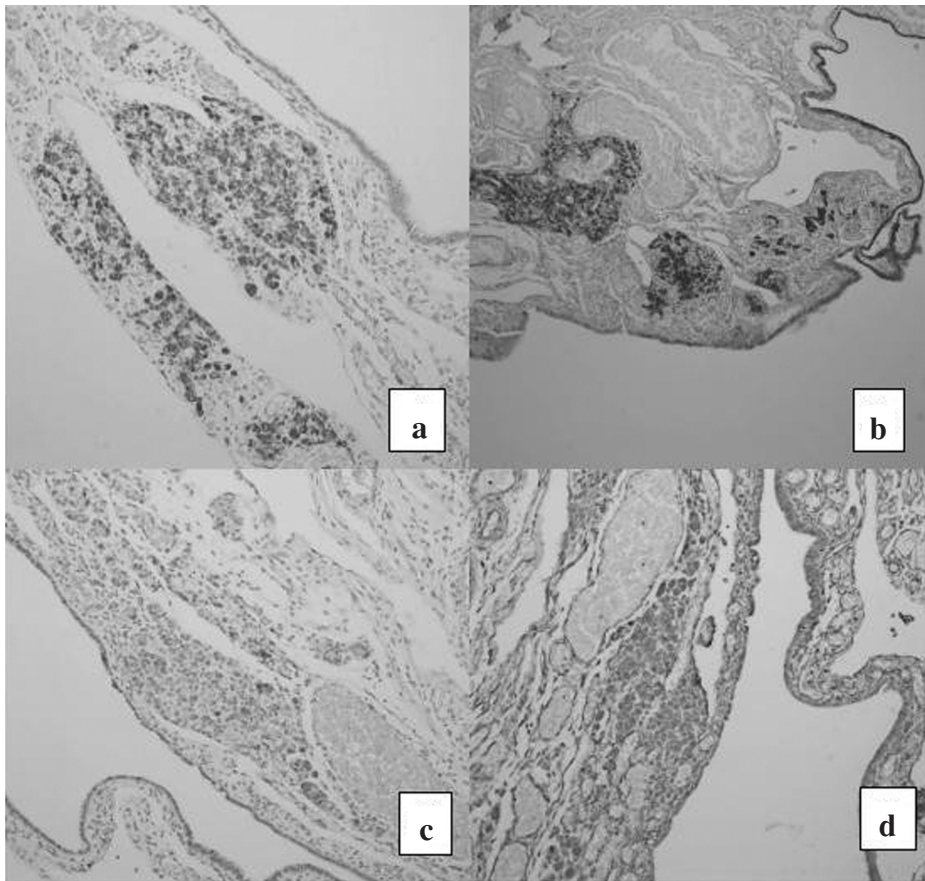


Figure 2. The hilus cells were immunohistochemically stained with; inhibin (a), calretinin (b), Melan-A (c), Vimentin (d).

Microscopic examination revealed a well differentiated endometrioid carcinoma of the endometrium, with more than one half infiltration of myometrium and without endocervical involvement (Figure 1D). Immunohistochemical findings were as follows; inhibin (1/25, Serotec), calretinin (1/100, NovoCastr), Melan A (1/100, Neomarkers), Vimentin (1/500, Neomarkers) were positive and EMA (1/100, Neomarkers), CD99 (1/100, Neomarkers) were negative (Figure 2).

DISCUSSION

Hilus cells are ovarian counterpart of testicular Leydig cells, with the exception of female chromatin pattern. Hilus cells are present during fetal life, not identified in childhood but reappear at puberty and are demonstrable in all

postmenopausal women (2). Hilus cell aggregates are typically found in the ovarian hilus. Few cases were reported with the existence of hilus cells in the fallopian tube (3,4). Apart from these case reports and another study detecting the presence of hilus cells in fallopian tube (5) no other article has appeared on that topic in the English written literature. Also, to our knowledge this appears to be the first report of an incidental finding of hilus cells in the fallopian tube coincident with an endometrial carcinoma.

The morphologic similarity of hilus cells with adrenal rest cells may be a diagnostic challenge in some cases. The presence of Reinke cyrystals may be a clue for the hilus cells, but immunohistochemical features are quite characteristic. Staining pattern of these hilus cells are similar to ovarian hilus cells and other sex-cord stromal tumors such as positive staining with in-

hibin, vimentin, melan A, calretinin and negative staining with EMA. In contrast to hilus cells, adrenal rests are usually positive with neuroendocrine markers. This staining characteristics also helps differentiate epithelial tumor from hilus cell aggregates. Epithelial tumors are usually positive with EMA and almost always negative with other markers.

To our knowledge this appears to be the first report of an incidental finding of hilus cells in the fallopian tube co-incident with an endometrial carcinoma. It is a well known fact that well differentiated endometrial carcinomas are associated with estrogens. Hilus cells are steroid hormone producing cells, although to what extent hilus cells contribute to the steroid hormone pool is unknown (1). The major product of hilus cells is the androstenedione, but small amounts of E2 and P are also produced (1). In a study of hysterectomy/oophorectomy specimens with non-neoplastic uterine lesions done in 1971 found that fifty two percent of fallopian tubes contain ectopic hilus cells (5). Although, there are some reports implying the association between hilus cell hyperplasia of the ovary and endometrial carcinoma (6), this is the first report implying the association between well differentiated endometrial carcinoma and extra-ovarian hilus cells.

The origin of hilus cells in the fallopian tube is a mystery. It is theorized that ovarian hilus

cells originated from undifferentiated ovarian mesenchyme, non myelinated nerve or perineural fibroblast. Lewis suggested that during embryologic development some ovarian mesenchymal cells might migrate into the mullerian ducts (3). Some others defined these lesions as heterotopia (5).

Whatever the origin, the presence of hilus cells in the fallopian tube is an incidental finding. Both microscopic and immunohistochemical features are identical to hilus cells of the ovary. So immunohistochemistry may be a useful tool to differentiate the extra-ovarian hilus cells from their mimics. Co-existence of the hilus cells in the tuba uterina of the patients having an endometrial carcinoma may be subject of further research.

REFERENCES

1. Clement PB. Anatomy and Histology of the Ovary, (In) Kurman RJ ed. Blaustein's Pathology of the Female Genital Tract 5th ed., New York, Springer Verlag, 2001. p.649-673.
2. Sternberg WH. The morphology and androgenic function, hyperplasia and tumors of human ovarian hilus cells. *Am J Pathol* 1949;25:493-521.
3. Lewis JD. Hilus-cell hyperplasia of ovaries and tubes: Report of a case. *Obstet Gynecol* 1964;24:728-731.
4. Palomaki JF, Blair OM. Hilus cell rest of the fallopian tube. A case report. *Obstet Gynecol* 1971;37:60-62.
5. Honore LH, O'Hara KE. Ovarian hilus cell heterotopia. *Obstet Gynecol* 1979;53:461-464.
6. Mohamed NC, Cardenas A, Villasanta U, Toker C, Ances IG. Hilus cell tumor of the ovary and endometrial carcinoma. *Obstet Gynecol* 1978;52:486-490.