

# Hilus Cell Hyperplasia of Fallopian Tubes: A Rare and Incidental Finding with Uterine Leiomyomas

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## Citation

Ansari IA, Prakash G, K R, V E, Bhat S. Hilus Cell Hyperplasia of Fallopian Tubes: A Rare and Incidental Finding with Uterine Leiomyomas. *Kathmandu Univ Med J* 2014;45(1):64-66.

## INTRODUCTION

Ovarian hilus cells are considered to be analogous with testicular Leydig cells with a female chromatin pattern.<sup>1</sup> Sternberg was apparently the first to employ the term ovarian hilus cells and stressed the likelihood of their being a source of androgen.<sup>2</sup> They are primarily found in the hilar region of the adult ovaries and rarely seen outside the ovaries especially the fallopian tubes.<sup>3-5</sup> Extensive search of literature showed only one case reporting co-existence of hilus cell hyperplasia of fallopian tube and uterine leiomyoma.<sup>4</sup> The following is a case report of incidental finding of hilus cell hyperplasia involving both the tubes in a postmenopausal woman with multiple uterine leiomyomata.

## ABSTRACT

Hilus cells are ovarian counterpart of testicular leydig cells seen at hilar region of ovary. We present an interesting case of hilus cell hyperplasia in fallopian tube associated with multiple leiomyomata of uterus in a postmenopausal woman. To our knowledge very few cases reported the existence of hilus cells outside the ovary. We received a specimen of hysterectomy with bilateral salpingo-oophorectomy. On gross examination both the tubes showed multiple small yellowish nodules. Microscopic examination showed aggregation of hilus cells beneath a normal tubal epithelium. Hilus cell hyperplasia was not seen in the ovaries. On immunohistochemistry, hilus cell showed positivity for vimentin and calretinin. Co-existence of hilus cell hyperplasia of fallopian tube and leiomyoma is very rare and may be subjected to further research

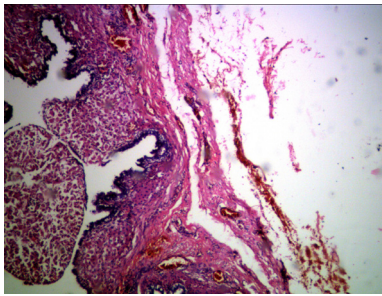
## KEY WORDS

*Fallopian tube, hilus cell hyperplasia, leiomyoma*

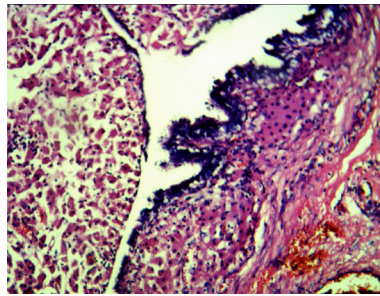
## CASE-REPORT

A 52 year old woman presented with chief complaints of a mass in the abdomen since two months. She had a past surgical history of cholecystectomy and appendicectomy done five years back. She attained menopause four years back and her menstrual history was normal. There was no history of any postmenopausal bleeding and any other complains.

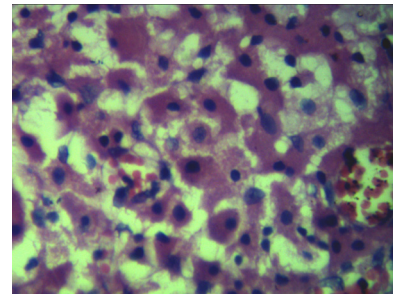
Examination of the abdomen revealed the presence of fibroid uterus along with incisional hernia. Gynaecological and radiological examination confirmed the clinical diagnosis of fibroid uterus. The provisional diagnosis of fibroid uterus and incisional hernia was made and the patient was subjected to total abdominal hysterectomy



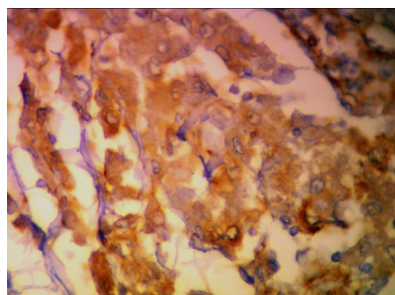
**Figure 1.** Photomicrograph of fallopian tube showing aggregates of large, polygonal cells with abundant granular eosinophilic cytoplasm & vesicular nuclei beneath the epithelium (H&E 40X).



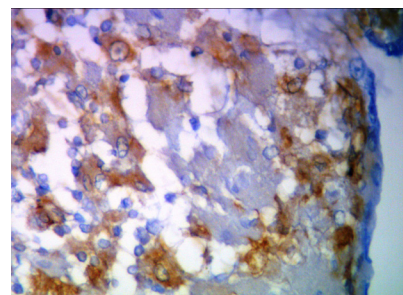
**Figure 2.** Photomicrograph showing aggregates of hilus cells beneath the epithelium (H&E 100X).



**Figure 3.** Photomicrograph showing a closer view of hilus cell aggregates with large, polygonal cells and abundant granular eosinophilic cytoplasm and vesicular nuclei beneath the epithelium (H&E 400X).



**Figure 4.** Photomicrograph showing vimentin positivity in hilus cells (IHC 400X).



**Figure 5.** Photomicrograph showing calretinin positivity in hilus cells (IHC 400X).

with repair of incisional hernia.

Specimen of hysterectomy with bilateral salpingoophorectomy was received. Uterus measured 14 x 8 x 8 cm and weighed 450 grams. Cut surface showed presence of multiple intramural leiomyomata of varying sizes, the largest measuring 4.5 cms in diameter and the smallest 1 cm in diameter. Endometrium was unremarkable. Both the tubes measured 5 cm in length. Right tube was dilated. Cut surface of right tube showed multiple yellowish nodules of varying sizes ranging from 1-3 mm. Left tube and both the ovaries were unremarkable.

On microscopic examination, endometrium showed proliferative pattern with focal areas showing simple hyperplasia, myometrium showed presence of leiomyomata. Ovaries showed normal histology. The interesting finding detected in fallopian tube was aggregates of hilus cells. The cells forming these aggregates were large, polygonal with abundant granular eosinophilic cytoplasm and vesicular nuclei. These aggregates of hilus cells were scattered throughout the stroma of the tubes beneath the normal epithelium. No prominent atypia or mitosis was found and Reinke's crystals were not seen (Fig 1,2 and 3).

These polygonal cells were further subjected to immunohistochemistry and showed positivity for vimentin (Fig 4) and calretinin (Fig 5) hence confirmed the presence of extra ovarian hilus cell hyperplasia in fallopian tubes.

## DISCUSSION

Ovarian hilus cells are morphologically similar to the Leydig cells of testes and are found in the fetal ovaries, then disappear during childhood and reappear at puberty. Larger nests or aggregates of hilus cells are found in majority of post-menopausal women and characteristically ensheath or lie within non medullated nerve fibres at the hilar region of ovary.<sup>3</sup> Very few reported cases showed the existence of hilus cells outside the ovary especially the fallopian tubes.<sup>4,5</sup> Apart from these case reports and another study detecting the hilus cells in fallopian tubes, no other article has appeared on this topic in the literature.<sup>5</sup> Also to our knowledge this appears to be the second report of an incidental finding of hilus cell in the fallopian tube associated with uterine leiomyoma. Lewis JD was the first author to report the coexistence of hilus cell hyperplasia in the tube with uterine leiomyom.<sup>3</sup> While, Alp Usbutun is the first person who reported one case of incidental finding of hilus cell hyperplasia in fallopian tube associated with endometrial carcinoma.<sup>1</sup>

The origin of ovarian hilus cell theorized to be from undifferentiated ovarian mesenchyme, non myelinated nerve or perineural or perivascular fibroblasts.<sup>1</sup> But the origin of hilus cells in the fallopian tube is a mystery. It is conceivable that during embryogenesis some ovarian mesenchymal cells might migrate into the mullerian duct.<sup>3</sup> Some authors had defined presence of extra ovarian hilus cell hyperplasia as heterotropia.<sup>5</sup>

The morphologic similarity of ovarian hilus cell to both luteinized stromal cells and to adrenal rest cells has been pointed out by several authors. Differentiation of hilus cells to the adrenal rest cells is sometime a great diagnostic challenge. The presence of Reinke crystals may be a clue for the hilus cells but it is not always a criterion to diagnose hilus cell and Reinke crystals are usually seen with tumor not with hyperplasia in such cases immunohistochemistry plays a vital role and is quiet characteristic. Hilus cells show positivity for inhibin, vimentin, melan A, calretinin and negative staining for EMA. In contrast adrenal rests are usually positive for neuroendocrine markers.<sup>3</sup> The distinction between extensive hilus cell hyperplasia and hilus cell tumor can be difficult, although a diameter of 1.0cm is frequently used as the minimum cut off for a tumor.<sup>6</sup>

Hilus cells are steroid hormone producing cells and the major product of hilus cells is androstenedione but small amount of Estrogen and Progesterone are also produced. Sternberg et al have produced evidences suggesting that increased gonadotrophin levels are the stimulus for development or hyperplasia of hilus cells.<sup>7</sup> However gonadotrophin studies were not done on this patient. Steroid secretion from hilus cells produced defeminization and masculinization in women. However no such features were seen in this patient and the only evidence of any

hormonal activity were the development of leiomyomata at a postmenopausal age and a focus of simple hyperplasia of endometrium.

## CONCLUSION

To the best of our knowledge very few cases have reported existence of hilus cells outside the ovary specially the tubes. The present case shows incidental finding of hilus cell hyperplasia in fallopian tubes with uterine leiomyomata. It is a well known fact that high production of steroid hormone especially estrogen is associated with endometrial carcinoma and some reports in the literature imply the association between hilus cell hyperplasia of ovary and endometrial carcinoma.<sup>8</sup> Hence extensive search of hilus cells in the tubectomy specimens with regular follow up of the patients for developing effects related to high estrogen can be a subject of further research.

## ACKNOWLEDGEMENT

We would like to thank Professor and Head, Department of Obstetrics and Gyneacology for providing the sample. We also express our gratitude to all technical and supporting staff for their constant help and support to complete this study.

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