



## Ichthyosis Uterus – A Rare Premalignant Lesion of Endometrium

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### ABSTRACT



Ichthyosis (ikTHEosis) uterus is a rare benign metaplastic transformation of uterine endometrium into stratified squamous epithelium. The word ichthyosis (plural ichthyoses) comes from two ancient Greek (ixtus), meaning fish and disease. The metaplastic epithelium is parakeratotic with absent granular layer. Premalignant potential of this benign transformation has not been established due to the rarity of occurrence. We report a case of 72 year old female, post menopausal with the complaints of painless lower abdominal distension for past six months. She had no history of menorrhagia or dysmenorrhea in her previous menstrual cycles. USG abdomen was done. She was taken up for elective hysterectomy with a diagnosis of hydrometra. Total abdominal hysterectomy with bilateral salpingo-oophorectomy with pelvic lymph node sampling done. Peritoneal fluid cytology, peritoneal biopsy was done. Gross section of specimen was seen and the same sent for histopathological examination. Based on these histological findings a diagnosis of Ichthyosis uteri with focal High grade Squamous intraepithelial lesion of endometrium was made. In our case, there is a direct continuity of the squamous metaplasia of the endocervix and the endometrial lining. This supports the benign metaplastic spread of squamous epithelium from the transformation zone of the cervix into the uterine cavity. However, the stimulation of squamous metaplasia in uterine endometrium as a result of long term hydrometra is being hypothesised in our patient. The association of hydrometra, squamous metaplasia and dysplastic changes in endometrium are speculated.

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### INTRODUCTION

A 72 year old, postmenopausal lady presented with the complains of painless lower abdominal distension for past six months. She had attained

menopause 25 years ago. She had no history of vomiting, diarrhoea, dysuria or fever and she had no history of menorrhagia or dysmenorrhea in her previous menstrual cycles.

On abdominal examination a nontender midline suprapubic mass of 10 X 12 cm arising from pelvis was felt. On per speculum examination the cervix was atrophied and stenosed. On per vaginum examination a uterine mass occupying all fornices was felt. On per rectal examination, there were no nodules in pouch of douglas.

Ultrasonography of upper abdomen and pelvis revealed a dumble shaped cystic mass measuring 162 X 100 X 110 cm posterior to the bladder.

There were no septations or internal echoes. Bilateral ovaries could not be visualised as separate.

**Case Report**

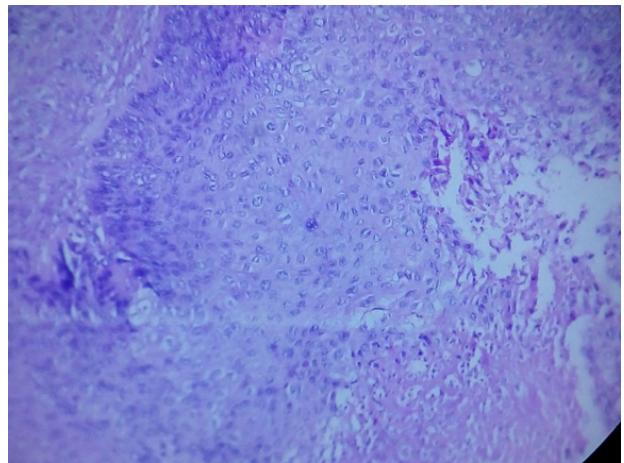
She was taken up for elective hysterectomy with a diagnosis of hydrometra. A suprapubic transverse incision was made.

Intraoperatively the uterus was enlarged. The cervix was also enlarged. Bilateral ovaries were atrophic. Peritoneal fluid cytology, peritoneal biopsy, total abdominal hysterectomy with bilateral salpingo-oophorectomy with pelvic lymph node sampling was performed. The specimen was sent for histopathological examination. The gross examination of the cut section showed widened endometrial cavity and endocervical canal filled with straw coloured clear fluid. The endometrial and endocervical surface were tan pink. Flattened and glistening. Both tubes and adnexa were unremarkable (Figure 1, Figure 2). In Figure 2, the cavities were filled with clear straw coloured fluid. There were no suspicious areas of dysplasia.

endometrium was replaced by stratified squamous epithelium with a single focus of high grade squamous intra-epithelial dysplastic changes (Figure 3, Figure 4). The epithelium lacked a granular layer and was parakeratotic. There was a single focus of nuclear hyperchromasia, mutinucleation, nuclear membrane irregularity and increased nucleocytoplasmic ratio (Figure 3). There was loss of cell stratification without breach of epithelial basement membrane. Extensive sampling did not reveal any endometrial glands or endocervical glandular cells. Cervical histopathology was normal. Based on these histological findings a diagnosis of Ichthyosis uteri with focal High grade Squamous intraepithelial lesion of endometrium was made.



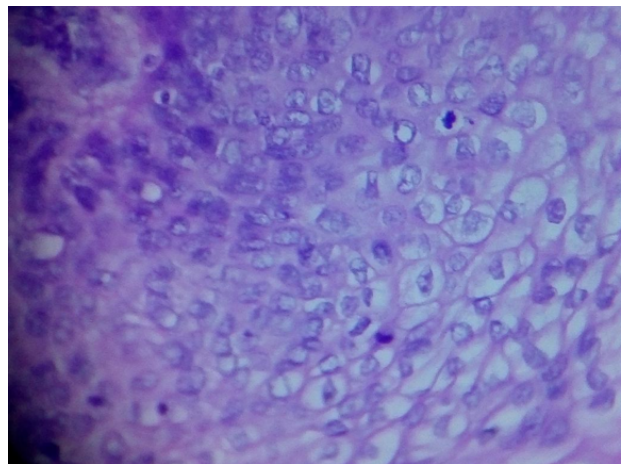
**Figure 1: Gross Appearance with Distended Uterus and Cervix**



**Figure 3: Haematoxylin and Eosin 100 X Showing Squamous Epithelium Lining the Endometrium with Underlying Thinned Myometrium**



**Figure 2: Cut section with tan pink, flat and glistening endometrium and endocervical canal**



**Figure 4: Haematoxylin and Eosin 400 X Squamous Epithelium with High Grade Dysplastic Intraepithelial Changes**

On microscopic examination the full thickness of

## DISCUSSION

The word ichthyosis (plural ichthyoses) comes from two ancient Greek (ixtus), meaning fish and disease [1]. Ichthyosis uteri is an extremely rare condition when the entire surface of endometrium is replaced by keratinized squamous epithelium. The origin of the term "Ichthyosis uteri" dates back to 1885 when it was first used by Zeller to describe the squamous metaplasia of entire surface of endometrium following introduction of caustic substances like iodine and formalise into the uterine cavity [1]. The aetiology was not well understood and was attributed to the inflammation caused by chronic irritation by a foreign material. The etiological role of Human papillomavirus was also investigated subsequently [2-4].

In our case, there is a direct continuity of the squamous metaplasia of the endocervix and the endometrial lining. This supports the benign metaplastic spread of squamous epithelium from the transformation zone of the cervix into the uterine cavity. However, the stimulation of squamous metaplasia in uterine endometrium as a result of long term hydrometra is being hypothesised in our patient.

The spread from the reserve cells of transformation zone is plausible but the de novo metaplastic transformation of endometrial glandular reserve cells into squamous cells is also likely. There is a possibility of reserve cells in endometrial basement membrane undergoing metaplastic transformation into squamous cells.

Literature review suggests that cases of dysplastic ichthyosis like changes are seen in association with squamous cell carcinoma of cervix [5-7]. It was concluded recently that significant cervical pathology should be ruled out whenever plaques of squamous epithelium or low grade dysplastic changes are seen in endometrial biopsy. Our case militates this idea because the uterine squamous epithelium was metaplastic in entirety with focal dysplastic changes in the absence of a coexisting cervical pathology. We believe that de novo ichthyosis uteri secondary to hydrometra is an independent risk factor for uterine squamous carcinomas. Points favouring this hypothesis is

1. The cervix showed normal stratified squamous epithelium.
2. The endometrium and endocervical canal were entirely metaplastic
3. The deepest point of invasion of dysplastic cells was in the endometrium and not cervix.

4. The extensive sampling did not reveal any endometrial or endocervical glandular cells.
5. In our case, the absence of koilocytic changes in the squamous metaplasia also excluded the Human papilla virus infection. Our case resembles earlier case reports where ichthyosis uteri was reported in the absence of cervical pathology [8-10]. As in these case reports, the probability of lepidic surface extension of cervical metaplastic reserve cells is unlikely the probability of reserve cells in endometrium that undergo benign metaplastic transformation can be speculated.

The role of Human Papilla Virus in uterine adenocanthomas is also well established though the role of Human papillomavirus in ichthyosis is unknown [4, 11]. Ichthyosis uteri has also been reported as a precursor of adenocarcinomas of the corpus uteri [12, 13].

## CONCLUSION

We believe that de novo ichthyosis uteri secondary to hydrometra is an independent risk factor for uterine squamous carcinomas. Long term hydrometra resulting from senile atrophic cervical stenosis in our patient could have induced reserve cell squamous metaplasia in endometrial and endocervical canal. The role of Human papillomavirus in Ichthyosis uteri is yet to be established.

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## Conflict of Interest

The authors declare that there is no conflict of interest.

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