

Icthyosis Uteri with in Situ Carcinoma - Report of 2 Cases

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ABSTRACT

Icthyosis uteri is a rare condition in which entire surface of the endometrium is replaced by stratified squamous epithelium. Two cases of Icthyosis uteri are reported for their rarity. The first case was of a 70 year old female with a history of lower abdominal pain and white PV (per vaginal) discharge since 3 months. The second case was of a 70 year old female with history of PV (per vaginal) spotting and breathlessness since 1 year. Gross examination of uterine cavity showed rough and irregular endometrial surface in the first case while in second case endometrium was atrophic. Microscopy of endometrium in both cases revealed total replacement of endometrial glands by stratified squamous epithelium with changes of in situ carcinoma or severe dysplasia. Cervix also showed severe dysplasia or CIN (cervical intra epithelial neoplasia) III in both the cases.

Keywords: *Dysplasia, CIN, pyometra, Hysterectomy*

Introduction

Icthyosis uteri is a wide spread replacement of endometrial surface by stratified squamous epithelium. It is a benign condition and may be associated with malignancy. We report 2 cases of ictyosis uteri with in situ carcinoma. Cervix show severe dysplasia or carcinoma in situ (CIN III) in a pan hysterectomy specimens in both elderly post-menopausal women. In second case we observed both fallopian tubes in their proximal parts show squamous metaplasia with severe dysplasia. We report these two cases due to their rarity.

Case Report

The first case was of a 70 year old post-menopausal female P3L3 presented with lower abdominal pain and white PV discharge since 3 months. Us was drained and cervical biopsy taken which was reported as cervical intra epithelial neoplasia grade III (CIN III) and hysterectomy was planned.

USG pelvis and TVS showed no significant abnormality. On CT abdomen there was bulky and heterogeneously enhancing cervix likely suggestive of neoplastic etiology.

The post hysterectomy specimen was sent for histopathological examination measured 9x7x3cms. Cut section revealed rough and irregular endometrial cavity (Fig.1). No ulceration or growth seen on cervix. Bilateral adnexa grossly appeared unremarkable.

The second case was of a 70 year old postmenopausal female P4L3 with history of PV spotting and breathlessness since 1 year. USG abdomen and pelvis showed bulky cervix. Endometrium is thin. Few intra cavity echogenic

contents are seen possibly due to small intra uterine collection. On CT abdomen cervix appeared bulky. It showed heterogenous enhancement. Uterus was atrophic. Endometrial collection measured 18x15x6 mm.

Cervical biopsy was reported as cervical intra epithelial neoplasia grade III (CIN III) . we received hysterectomy specimen with bilateral adnexa measured 5.5x4.5x2 cms. Fallopian tubes were 4.5 cms in length and ovaries measured 2x1 cms each. Uterus was atrophic. No ulceration or growth seen on cervix.

Microscopically endometrial cavity in both the cases was lined by squamous epithelium showing features of carcinoma in situ. (Fig.2) At places keratinization seen. No invasive focus of squamous cell carcinoma noted into the myometrium. Ectocervix and endocervix were lined by stratified squamous epithelium showing severe dysplasia or CIN III. Both the fallopian tubes of the second case in their proximal part were lined by squamous epithelium showing carcinoma in situ (Fig.3) while middle and distal parts were unremarkable. Bilateral ovaries were unremarkable in both the cases.

Discussion

The term Icthyosis uteri refers to extensive squamous metaplasia of surface endometrium following iatrogenically introduced caustic substances such as formaline or iodine was first coined by Zeller in 1885.^[1] This rare pathology has been seen in association with conditions like tuberculous endometritis, puerperal endometritis, endometrial polyps, hyperplasia, squamous papilloma and pyometra as a result



Fig. 1: Cut section of uterus with rough and irregular endometrial cavity..

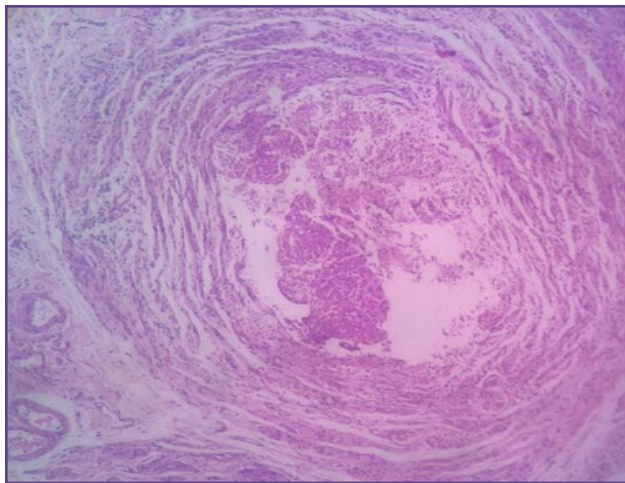


Fig. 3: Lumen of the fallopian tubes in their proximal parts were lined by squamous epithelium showing carcinoma in situ. (H&E stain, x100)

of cervical stenosis or malignancy^[2] The average age of presentation of this condition is 50 to 70 years as described by various authors and one case described where the patient's age was 38 years who was immunocompromised. ^[3] we reported 2 cases of ichthyosis uteri in a span of one year where total number of histopathological specimens received were 5572 . Squamous cell carcinoma of endometrium may arise either directly or in association with ichthyosis uteri. ^[4]Two authors described ichthyosis uteri along with endometrial adenocarcinoma . ^[2,5] The first case described here in showed extensive ichthyosis uteri of endometrium showing features of severe dysplasia with history of pyometra along with CIN III. Pyometra was reported by other authors as an associated factor. ^[4,6,7] The second case described with ichthyosis uteri of endometrium

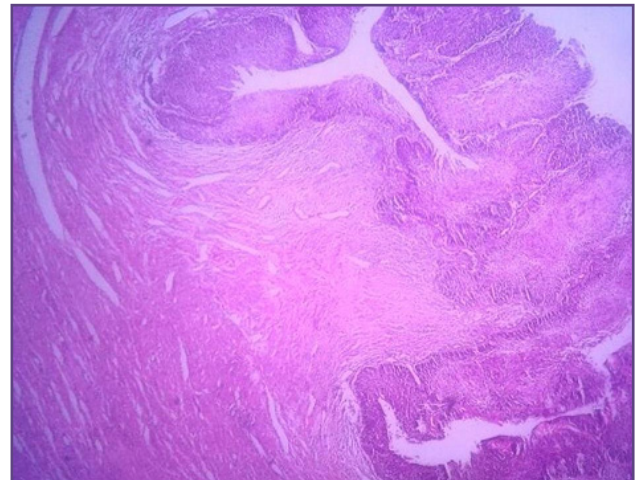


Fig. 2. Microscopy of endometrium showed total replacement of endometrial glands by stratified squamous epithelium with changes of carcinoma in situ or severe dysplasia. (H&E stain, x100).

showing carcinoma in situ with CIN III. Proximal parts of fallopian tubes show squamous metaplasia with in situ carcinoma. One of the author described ichthyosis uteri with involvement of fallopian tube. ^[4] The squamous metaplasia of the fallopian tube could be on account of proximal extension from extensive ichthyosis in the endometrium. ^[4]

Factors which favour this condition in our both cases are thickened and rough endometrium showing severe dysplasia or carcinoma in situ, no obvious growth in cervix and postmenopausal age of the patients. ^[4]

These findings may be explained in two ways. The first explanation is squamous cell carcinoma originated in the cervix and extended proximally colonizing pre-existing ichthyosis uteri. The second explanation is lipedic surface extension of a cervical squamous cell carcinoma with no pre existing ichthyosis uteri. ^[8] The etiology of endometrial keratinisation is not well understood. Chronic trauma, repair, irritation, foreign material and estrogenic effects have all been implicated. ^[2,9]

Conclusion

Ichthyosis uteri is a rare condition showing squamous metaplasia of endometrium leading to development of squamous cell carcinoma of endometrium.

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Financial or other Competing Interests: None.