Case Report:
A Rare Case of Primary Insitu Squamous Cell Carcinoma of the Endometrium with Extensive Icthyosis Uteri.

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Abstract: Primary squamous cell carcinoma of the endometrium is exceedingly rare. We report a case of 52 years old postmenopausal woman who presented with pelvic pain of four months duration. Gynecologic examination revealed a normal cervix. A possibility of pyometra was considered through pelvic ultrasound. Total abdominal hysterectomy was performed and histopathologically, it was diagnosed as a case of primary in situ squamous cell carcinoma of the endometrium.

Key Words: Dysplasia, Endometrium, Pyometra, Squamous cell carcinoma.

Introduction: Primary squamous cell carcinoma of the endometrium is an extremely rare entity. It has been described in association with pyometra, cervical stenosis, chronic inflammation, multiparity and ichtyosis uteri and is usually seen in postmenopausal women. ichtyosis uteri is a rare condition in which the entire surface of endometrium is replaced by stratified squamous epithelium.(1) Primary in situ squamous cell carcinoma arising in ichtyosis uteri is still rare with a very few case reports in the literature.

Case Report:
A 52 years old lady presented with pelvic pain since four months. She was para 3, had 3 living children and had attained menopause 10 years back. She had no vaginal bleeding or iatrogenically introduced substance in the uterus in the past. General Physical Examination was unremarkable. Gynecological examination revealed a normal cervix. A pelvic ultrasound was performed and showed an enlarged corpus with accumulation of liquid within the endometrial cavity. Hence a presumptive diagnosis of pyometra was made. A cytological examination of fluid collected in the endometrial cavity revealed a few dysplastic squamous cells. Hence, total abdominal hysterectomy with bilateral salpingoophorectomy and removal of right and left pelvic lymphnodes was done.

Pathological findings:
The uterus with cervix measured 9 x 6 x 2 cm. Cut surface showed widened endometrial cavity lined by irregular, shaggy, membrane like material (Fig.1) Myometrium and bilateral adnexae were unremarkable. Cervix showed focal areas of erosion. Multiple sections from endometrium showed replacement of the entire endometrium by stratified squamous epithelium with extensive areas of moderate dysplasia and carcinoma in situ (Fig 2,3). Only occasional atrophic endometrial glands were seen covered by areas of ulceration(Fig 4). Underlying myometrium was unremarkable. Cervix was unremarkable except for focal areas of ulceration. There was no evidence of malignant glandular tissue upon sectioning the entire endometrium. Hence, a final diagnosis of primary squamous cell carcinoma of endometrium was contemplated.

Fig 1: Widened endometrial cavity and shaggy membrane like material
Discussion

Zeller in 1985 first coined the term “ichthyosis uteri” in which the entire surface of endometrium is replaced by stratified squamous epithelium following, iatrogenically introduced substances like formalin or iodine.(1-3) The presence of squamous epithelium in the endometrium is variously termed ichthyosis uteri, leukoplakia, epidermalization psoriasis uteri, epidermoid heteroplasia, cholesterometra, acanthosis and indirect regenerative squamous metaplasia.(1-4) Chronic endometritis, hypovitaminosis A, estrogen deficiency, senile involution, irradiation, chronic irritative processes such as intrauterine devices are considered pathogenetic factors of squamous metaplasia of endometrial glands. Chronic irritation due to long standing pyometra might have lead to the malignant change in ichthyosis uteri.

According to Zains et al, ichthyosis lack malignant potential. However, dysplastic and anaplastic changes have been reported. Goodman found only six cases of squamous cell carcinoma of endometrium amongst 1182 cases of uterine corpus tumor.

Various studies have put forth two plausible explanations for the occurrence of squamous cell carcinoma of endometrium. Whereas one theory is that the direct extension of squamous cell carcinoma of cervix to the endometrium, other is its development from reserve cells. In the present case, since cervix did not show any gross or microscopic abnormality, the possibility of direct extension from squamous cell carcinoma cervix was ruled out.

To be accepted as a primary squamous cell carcinoma of endometrium, the tumor must satisfy the criteria established by Fluhmann and modified by Kay. There must be no coexisting endometrial adenocarcinoma; there must be no connection between endometrial tumor and squamous epithelium of cervix; there must be no squamous cell carcinoma of cervix; and if cervix shows an in situ carcinoma, there must be no connection between this and independent endometrial neoplasm. Our patient satisfied these criteria.(2-7)

The presence of spectrum of changes from squamous metaplasia through dysplasia to carcinoma in situ favored the second theory. The therapy consists of Total Abdominal Hysterectomy with bilateral adnexectomy followed by radiotherapy in selected cases. Chemotherapy can be considered in addition to radiotherapy in these patients. In endometrial carcinoma, presence of a malignant squamous cell component worsens the prognosis.(3-6) Prognosis is stage dependent, one fourth die within two years of diagnosis. Survival rate for patients with stage I disease is 80% and for stage II, it is 20%.(3-7) Hence early diagnosis and prompt diagnosis is imperative to improve the survival rate.

Although rare, possibility of diagnosis of primary squamous cell carcinoma of endometrium should be considered in a postmenopausal elderly females presenting with pyometra.

References