Case Report:
Squamous Cell Carcinoma of Endometrium with Extensive Icthyosis Uteri

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Abstract:
We report a rare case of squamous cell carcinoma of endometrium arising in icthyosis uteri in a 60 years old lady presenting with vaginal bleeding.

Key Words: Squamous cell carcinoma; Endometrium; Icthyosis uteri; Vaginal bleeding

Introduction:
Icthyosis uteri is a rare condition in which the entire surface of endometrium is replaced by stratified squamous epithelium. Squamous cell carcinoma of endometrium (SCC E) arising in icthyosis uteri is still rare, with only few cases reported in literature.

Case Report:
A 60 year female presented with history of bleeding per vaginum since 6 to 8 months duration. She was para 3, live 3 and had attained menopause 15 years back. There was no history of tuberculosis or irogenically introduced substance in the uterus in past.
Per speculum examination showed an atrophic cervix which was flushed with vagina.
Per vaginal examination revealed an enlarged uterus of 14 weeks gestation. Adnexa was not palpable through the cervix. Cervix appeared unremarkable.
Ultrasonography revealed large fluid collection in the uterine cavity along with small soft tissue projections over the margins of cavity.
Hence a presumptive diagnosis of pyometra was made. Endometrial cavity was drained by cervical dilatation. Endometrial curettage was done, histopathologic examination of which revealed strips of stratified squamous epithelium with high grade dysplasia. No endometrial glands or stroma was identified.
At surgery radical hysterectomy with adnexectomy was done.
Gross specimen consisted of uterus with cervix measuring 12 x 11 x 7 cm. Cut surface showed widened endometrial cavity lined by irregular shaggy, membrane like material. Also seen were grey white irregular nodules in the myometrium. Cervix was unremarkable.

Figure 1: Widened endometrial cavity and an irregular mass in the myometrium.

Multiple sections from endometrium showed replacement of the entire endometrium by stratified squamous epithelium which showed transition from dysplasia through carcinoma in situ, area of micro invasion and frankly invasive squamous cell carcinoma in the myometrium. Cervix was unremarkable. Final diagnosis of primary SCC-E with extensive icthyosis uteri was made.
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. It usually occur secondary to chronic inflammation, irritation, foreign bodies or following iatrogenically introduced substances in to the uterine cavity. According to some authors ichthyosis lack malignant potential. However, dysplastic & anaplastic changes have been reported, with few exceptional case reports describing the occurrence of primary SCC-E in the background of ichthyosis uteri. Till date only 64 cases of SCC-E have been reported in the literature. Among 1182 cases of uterine corpus tumor, Goodman found only six cases of SCC-E. Various studies have put forth two plausible explanations for the occurrence of SCC-E. One theory being the direct extension of squamous cell carcinoma of cervix to the endometrium, other being 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. Also the presence of spectrum of changes from squamous metaplasia through dysplasia, carcinoma in situ, microinvasion & frankly invasive squamous cell carcinoma favoured a primary SCC-E rather than extension from cervix which in turn will not show such changes.

Chronic irritation due to long standing pyometra might have lead to the malignant change in ichthyosis uteri.

Fluhmann and Kay had put forth following criteria for the diagnosis of primary SCC-E. 1) there should be no endometrial carcinoma, 2) no primary squamous cell carcinoma of cervix and 3) if there is in situ change in cervix there should be no association with the endometrial neoplasm. Our case satisfied all the above criteria.

Therapy consists of total abdominal hysterectomy with adnexectomy followed by radiotherapy in selected cases. Chemotherapy can be considered in addition to radiotherapy in these patients. Prognosis is stage dependent, one forth die within two years of diagnosis. Survival rate for patients with stage I disease is 80% and for stage III it is 20%. Hence early diagnosis and prompt treatment is imperative to improve the survival rate.

In summary, though rare, primary squamous cell carcinoma of endometrium can arise in ichthyosis uteri. Hence the possibility of squamous cell carcinoma of endometrium should be considered when strips of stratified squamous epithelium are found in endometrial curettage specimen especially in a postmenopausal women with pyometra.

References: