



Case Report

Cervical intraepithelial neoplasm with ichthyosis uteri- A case report

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ABSTRACT

A rare condition in which the entire surface of the endometrium is replaced by stratified squamous epithelium is called Ichthyosis uteri. Originally described as an endometrial response to iatrogenically-introduced caustic substances, similar changes have since been described in association with a variety of inflammatory conditions of the endometrium.

Here we report a case of intraepithelial neoplasm III, with ichthyosis uteri. A 75-years-old female with hypetension, underwent total abdominal hysterectomy with bilateral salphingo-ophorectomy for watery discharge per vaginal since four months.

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INTRODUCTION

Cervical intraepithelial lesion (CIN) is a premalignant condition with abnormal growth of squamous cells on the surface of the cervix.¹ CIN remains stable in most of the cases, or eliminated by the host's immune system without intervention.² However, a small percentage of case progresses to become cervical cancer, usually squamous cell carcinoma (SCC), when left untreated.³ The risk of progression to invasive cancer increases with time and the time required is shorter with increasing severity of the lesion. Cervical squamous cell carcinoma can directly invade the uterine wall with or without parametrial involvement or by lymphatic invasion. Direct extension to the endometrium replacing it without myometrial invasion is very uncommon.⁴

Ichthyosis uteri, is an exceedingly rare condition in which the surface of the endometrium is completely or near-completely replaced by squamous epithelium. Originally described as an endometrial response to iatrogenically-introduced caustic substances, similar changes have since been described in association with a variety of inflammatory conditions of the endometrium.⁵ Here we report a case with

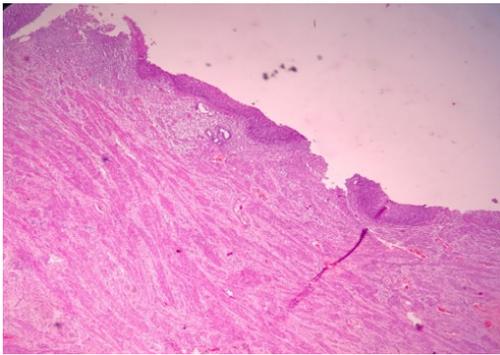


Figure 1: Replacement of endometrium lining by stratified squamous epithelium (HE stain, X40).

associated CIN 3 with surface of endometrium completely replaced by squamous epithelium.

CASE REPORT

A 75 years-old-female presented with watery discharge per vagina for four months and lower back pain from last 2 years. She had menopause 25 years back. There was no history of spotting, per vagina bleeding or lower abdomen pain. Her past history revealed hypertensive and arthritis for which she was under medication. No history of tuberculosis, inflammatory conditions of the uterus or iatrogenically introduced substances were found. On clinical examination she had eight week size anteverted firm uterus. Cervix showed sign of discharge with VIA positive on colposcopy with no obvious growth. The patient underwent a total abdominal hysterectomy, bilateral salpingo-oophorectomy. There was 30ml of pus in uterine cavity. The pus culture showed no growth. The procedure was well-tolerated without having any complications.

Gross examination: Specimen consisted of uterus with bilateral tubes and ovaries. Uterus measured 8× 6.5× 2.5cm. Endometrial cavity measured 3.7×0.5cm with tan to tan-brown area extending from cavity to endocervix and into the lower uterine segment. Endocervical canal measured 3.0cm in length and external os measured 0.6cm. Erosion was noted in cervix. Right tube measures 3.5×0.5cm and right ovary was cystic, measuring 2×1.5cm. Cut section showed cystic structure measuring 0.5cm, containing clear fluids. Left tube measured 4.5×0.5cm and left ovary measured 1.5×1.0×0.5cm, cut section showed greyish brown areas.

Microscopic examination: Sections from endometrium showed the entire endometrial surface is lined by non-keratinized stratified squamous epithelium revealing full-thickness dysplasia (fig.1). The endometrial gland and stroma beneath the surface were in proliferative phase. (fig.2) Section from cervix showed stratified squamous epithelium revealing loss of polarity and full thickness dysplasia with intact basement membrane (fig.3). The individual cells showed pleomorphism with high N:C ratio, enlarged irregular, hyperchromatic nuclei, prominent

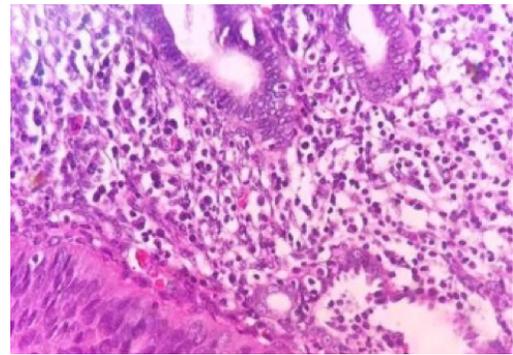


Figure 2: Stratified squamous epithelial lining showing features of dysplasia. Note the presence of endometrial glands within the stroma (HE stain, X100).

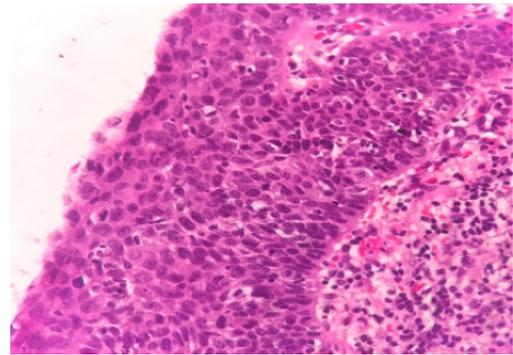


Figure 3: Stratified squamous epithelium of cervix showing full thickness dysplasia (HE stain, X100).

nucleoli. Mitotic figures were also seen up to middle third of epithelium. The underlying stroma showed chronic inflammatory cells consisting of lymphocytes. Normal looking endocervical glands were also noted. The section from right ovary showed cystic structures lined by benign cuboidal epithelium resembling features of simple cyst.

DISCUSSION

The case reported here is a cervical intraepithelial neoplasm grade 3 associated with extensive ichthyosis uteri-like changes of the endometrium that, additionally, had superimposed high-grade dysplastic changes. A case of carcinoma in situ of cervix with contiguous extension to the endometrium of the lower uterine segment is reported by Rajeshwari K Muthusamy et. al in their study.⁶ This histology findings may be explained due to viral infection in the cervix by HPV which lead to, a cervical intraepithelial neoplasm grade 3 originated in the cervix and the extended proximally, ichthyosis uteri. Oluwole Fadare in their study shows HPV positive in ichthyosis uteri with squamous cell carcinoma.⁵

The term "ichthyosis uteri" was initially coined in 1885 by Zeller to refer to extensive squamous metaplasia of the surface endometrium following iatrogenically-introduced caustic substances such as formalin or iodine.⁷ Since that

initial report, the term "ichthyosis uteri" and the phenomenon it describes have become well accepted but have been used only sporadically in the literature.^{8,9} Squamous metaplasia, a normal physiological process, is encountered frequently in cervical biopsies and loop electrosurgical excision procedures. The metaplastic cells are generally uniform, round to oval, with a single nucleolus. The nuclear contours are generally smooth. The presence of significant cellular crowding, nuclear atypia and increased mitotic figures in the upper half of the epithelium can be the most helpful morphological features in supporting a diagnosis of high grade CIN.¹⁰ Here they have all these features extended up to the surface of endometrium. Ichthyosis is considered as benign condition. However, anaplastic and dysplastic changes have been reported by some authors.^{9,11,12}

A case reported by Oluwole Fadare in 2006 showed 38 years female with complain of vaginal discharge with past medical history of acquired immune deficiency and the age group was in concordance with our case. Nishu Bhardwaj et al¹³ and Kanchan Murhekar, Urmila Majhi, et al¹⁴ shows dysplastic features in older age group. Pins et al described a case in which high-grade dysplasia of the cervix extended proximally and coated the entire endometrium.¹² Bewtra et al¹⁵ described a case of extensive benign squamous keratinization with underlying endometrial adenocarcinoma. Besides the dysplastic or anaplastic changes, a case of malignant degeneration has also been reported in ichthyosis uteri.¹⁶ This case presented as a CIN III at the base of ichthyosis uteri. The ichthyosis uteri developed over a period of just three years from a discrete squamous cell metaplasia of the endometrial body in the presence of slight nonspecific endometritis. Our case closely resembles the case described by Heckerroth and Ziegler.¹⁶

Conflict of Interest: None

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