

A Case Report of Ichthyosis Uteri

Zhouying Liu

Beijing Hospital

Dongge Liu (✉ dgliubjyy@126.com)

Beijing Hospital

Case Report

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Abstract

Background: Ichthyosis uteri is a rare condition in which the entire surface of the endometrium is replaced by squamous epithelium. The condition often is considered as benign, despite anaplastic and dysplastic changes have been reported.

Case presentation: A 36-year-old female present with extensive squamous metaplasia and dispersed normal endometrium. The case displayed with a benign ichthyosis uteri showing heavy keratinization without dysplasia. The cervix showed no dysplastic changes without HPV infection.

Conclusion: We conclude that a benign ichthyosis uteri was not associated with HPV infection.

Background

Ichthyosis uteri is an exceedingly rare condition in which the entire surface of the endometrium is replaced by stratified squamous epithelium. It was initially reported in 1885 by Zeller. Originally described as an endometrial response to iatrogenically-introduced caustic substances such as formalin or iodine, similar changes have since been described in association with a variety of inflammatory conditions of the endometrium.

Progesterone is a steroids released by ovary. As a sex hormone, it is applied to prevent miscarriage, emergency contraception et al in clinical practice. We describe herein an undescribed example of an ichthyosis uteri following after oral administration of progesterone.

Case Presentation

A 36-year-old female presented with complaints of massive vaginal bleeding for ten days. Her past medical history was insignificant with no history of tuberculosis, inflammatory conditions of the uterus or iatrogenically-introduced substances in uterus.

Laboratory investigations revealed hemoglobin (Hb) value was 96g/L (Normal Hb 115-150g/L).

Ultrasound abdomen (Fig. 1) showed endometrial thickness of 1.2 cm. Hysteroscopy examination showed uterus were significantly enlarged like a barrel, with endometrial cavity full filled with friable, grey-white mass. Cervical biopsy displayed stratified squamous epithelium without dysplasia and HPV infection. Endometrial curetting was grey-white and measured 3.5x3x1cm in size. Curettages were totally sampled and revealed stratified squamous epithelium showing heavy keratinization and no dysplastic changes (Fig. 2A, 2B). Extensive squamous metaplasia of endometrium and dispersed normal endometrium were seen (Fig. 2C) according to full sampling of endometrium and careful observation. P40, P16, P53, Ki-67 and CD10 were also detected by immunochemistry. P40 was extensively positive (Fig. 2D). Squamous epithelium was displayed with positive stain of p16 (Fig. 2E), wild-type p53 (Fig. 2F),

and low Ki-67 (5%, Fig. 2G). Endometrial stroma was found dispersed in squamous epithelium according to CD10 stain (Fig. 2H).

She was without any complications follow-up 9 months after her curettage.

Discussion & Conclusion

Zeller coined "ichthyosis uteri" in 1885 to describe extensive squamous metaplasia of the surface endometrium following iatrogenically introduced caustic substances such as formalin or iodine(1). The condition is exceedingly rare with few cases reported in the literature. Malignancies have rarely been associated with ichthyosis uteri. The case in the literature that most closely resembles our case was reported by Kucukali in 1996(2). The case described in that report underwent a benign papilloma with a background of severe ichthyosis uteri. Microscopic evaluation showed simulating koilocytosis in some papilloma cells without sign of dysplasia or malignancy. Human papillomavirus was not detected immunohistochemically.

Although ichthyosis is considered as a benign condition, anaplastic and dysplastic changes have been reported (3–5). These cases were associated with dysplasia or carcinoma of cervix. After discussion for direct extension from cervical for findings in endometrium. They conclude that squamous cell carcinoma of endometrium was either developed directly from(6) or in association with pre-existing ichthyosis uteri, and ichthyosis may associate with human papillomavirus(3).

A case of endometrial adenocarcinoma covered almost by a plaque-like, keratinizing mature squamous epithelium was described by Bewtra(7). Therefore, ichthyosis uteri could be diagnosed after extensive observation to exclude endometrial adenocarcinoma.

In summary, a case of a benign ichthyosis uteri without HPV infection is described. The possibility of squamous cell carcinoma should be considered after extensive examination (such as CEA in blood), especially when plaques of squamous epithelium with dysplastic changes are identified in an endometrial biopsy or curettage.

Abbreviations

HPV: Human papillomavirus; Hb: hemoglobin;

Declarations

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Authors' contributions

Zhouying Liu carried out concepts & design, data acquisition, data analysis & manuscript preparation. Lan Chen and Dongge Liu carried out concepts and literature search. All the authors have read & approved the final manuscript.

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Availability of data and materials

All the data regarding the findings are available within the manuscript.

Ethics approval and consent to participate

Not applicable.

Consent for publication

Written consent for publication and any additional related information was taken from the patient involved in the study.

Competing interests

The authors declare that they have no competing interests.

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Figures

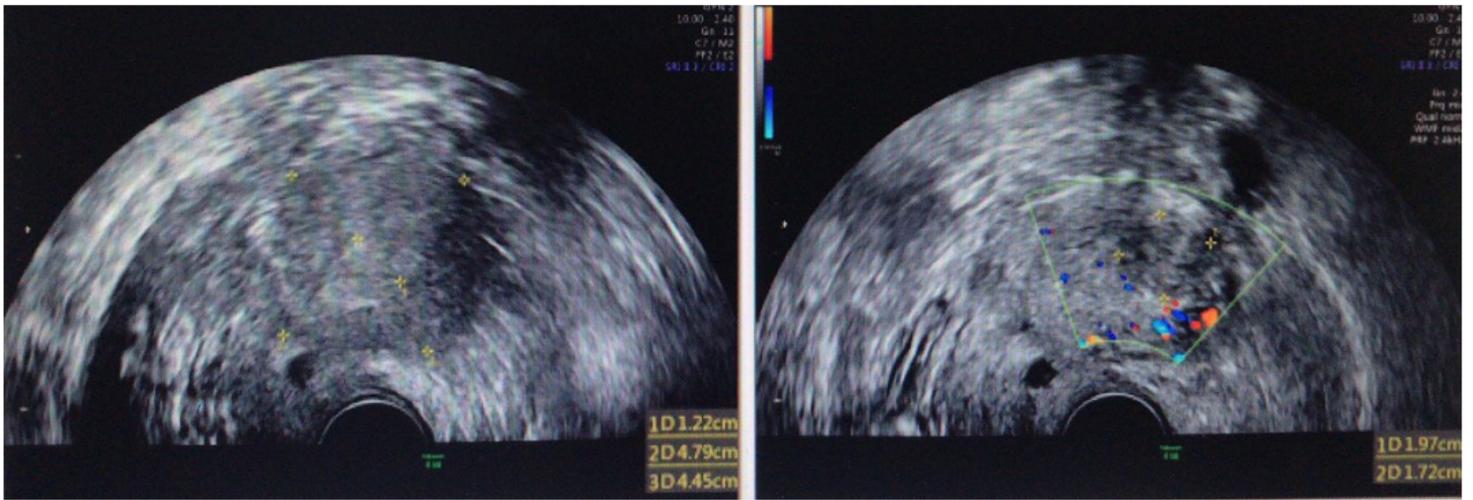


Figure 1

Ultrasound abdomen showed endometrial thickness of 1.2 cm.

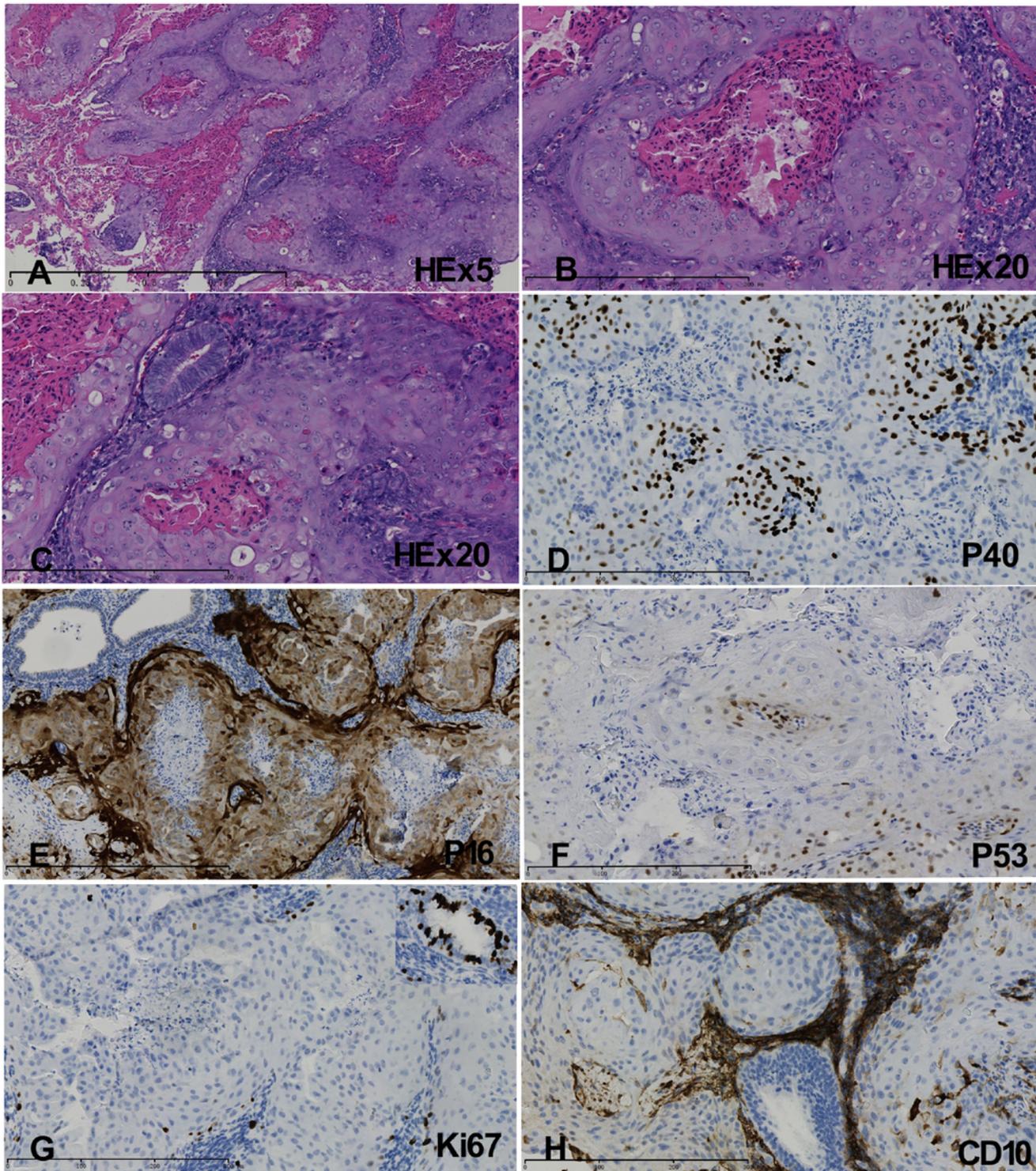


Figure 2

Squamous epithelium arranged in papillary pattern showing heavy keratinization and no dysplastic changes. A-C: Extensive squamous metaplasia of endometrium overlying myometrial stroma. D-H: immunochemistry stain of P40, P16, P53, Ki-67 and CD10 to show squamous epithelium with positive stain of p16, wild-type p53 and low Ki-67. CD10 display myometrial stroma.

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