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LETTER TO THE EDITOR

Adenoid Basal Carcinoma of the Uterine Cervix in Association with Keratinizing Squamous Cell Carcinoma: a Rare Diagnosis

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Editor,

Adenoid basal carcinoma (ABC) is an unusual cervical tumor which is considered to be indolent in nature when pure [1]. Detected incidentally in postmenopausal women, pure ABCs do not metastasize [2]. Hence, it is important to distinguish ABC from aggressive cervical tumors like adenoid cystic carcinoma and basaloid squamous cell carcinoma. However, ABC can be found co-existing with a higher-grade cervical malignancy which alters the prognosis and treatment for these patients [3, 4].

A 65-year-old postmenopausal lady presented at the obstetrics and gynecology department with complains of foul smelling, blood tinged vaginal discharge for 15 days. She was gravida 5, parity 5 with no significant personal or family history.

On per speculum examination, a 3 × 3 cm growth covering both anterior and posterior cervical lips was seen which bled on touch. The bilateral adnexa, parametrium, and rectal mucosa were free on palpation. This was followed by a punch cervical biopsy which showed adenoid basal carcinoma (Fig. 1a). The tumor was clinically staged as T_2a_2, and Wertheim's hysterectomy with pelvic lymph node dissection was performed.

Formalin fixed-gross specimen of the uterus and cervix with bilateral fallopian tubes together measured 9 × 7.5 × 2.6 cm. An exophytic growth was seen arising from the lower end of the cervix measuring 4 × 3 × 2.6 cm (Fig. 1b). The cut surface of the tumor was gray white in color and showed focal areas of necrosis and hemorrhage. The uterine body, bilateral fallopian tubes, adnexa, vaginal cuff, and parametria were unremarkable on gross inspection. Left pelvic, left obturator, and right external iliac lymph nodes were submitted and were grossly unremarkable.

On microscopy, the tumor showed all the four distinct tumor components of ABC described by Cviko A et al.: (i) a squamous invasive component (Fig. 2b), (ii) basal cells arising from squamous areas (Fig. 2c), (iii) few areas showing glandular differentiation (Fig. 2d, e), and (iv) high grade squamous intraepithelial lesion (Fig. 2a) [5]. Along with this, there were areas of invasive keratinizing squamous cell carcinoma (Fig. 2f). These areas constituted approximately 40% of the tumor. Alcian blue positive secretions were noted in the glandular component (Fig. 3a). Ki 67 staining showed a very high labelling index (40%) in the squamous cell carcinoma areas (Fig. 3c) while it was negative or very weak in the glandular or adenoid areas (Fig. 3d). However, an occasional focus of adenoid pattern showed an intermediate labelling index (15%) (Fig. 3b). The basaloid tumor cells were negative for synaptophysin and chromogranin. The vaginal cuff was microscopically free of tumor. Sections from the uterus, parametrium, bilateral fallopian tubes, and ovaries were histologically unremarkable. All the submitted nodes were free of tumor and showed reactive lymphadenitis. On the 14th post-operative day, the patient developed deep vein thrombosis and expired on the 39th-day post-surgery possibly due to pulmonary embolism/myocardial infarction.

Morphologically, ABC can be confused with adenoid cystic carcinoma (ACC), both of which are associated with cervical dysplasia and present in postmenopausal women [6]. However, careful attention to the morphology of the tumor cells can help differentiate the two entities. ACC shows larger nests of tumor cells, more commonly show cribriform arrangement, and the tumor cells show higher mitotic rate and nuclear pleomorphism. Tumor nests in ACC also elicit a significant stromal reaction [6].

Cervical adenoid basal carcinoma (ABC) can rarely be associated with conventional cervical malignancies like invasive squamous cell carcinoma (keratinizing or non-keratinizing), adenoid cystic carcinoma, and small cell neuroendocrine

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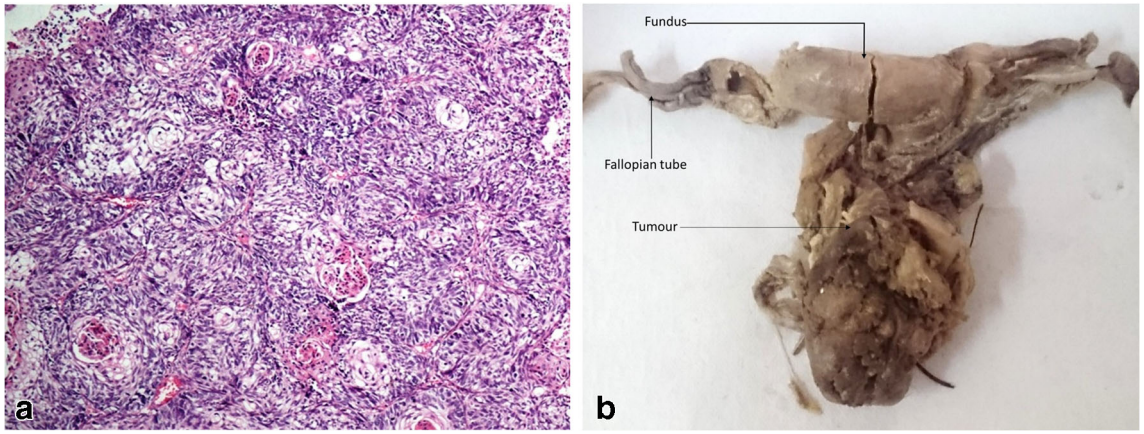

Fig. 1 **a** Biopsy cervix (hematoxylin and eosin, 40x)—tumor composed of small, uniform basaloid cells arranged in small nests with rounded appearance and central area of squamous differentiation. **b** Gross specimen of the uterus and cervix with bilateral fallopian tubes. An exophytic growth seen arising from the lower end of cervix

carcinoma and carcinosarcoma [3, 4]. This can markedly alter the prognosis and treatment modality, making an adequate biopsy essential to provide a clear diagnosis and therapeutic plan. The presence of a coexistent malignancy in ABC and presence of squamous differentiation and glandular structures in pure ABC can be explained by the hypothesis that ABC arises from the cervical reserve cell population which can show multidirectional differentiation [3]. The present case showed typical morphological features of ABC along with the presence of an invasive squamous cell carcinoma (keratinizing type). The Ki 67 labelling pattern further highlights the distinct components within the same tumor [7]. The higher labelling index in the squamous cell carcinoma areas points to a poorer prognosis.

In conclusion, adenoid basal carcinoma can be associated with a higher-grade cervical malignancy which warrants a careful inspection as it can markedly alter the prognosis and treatment plan.

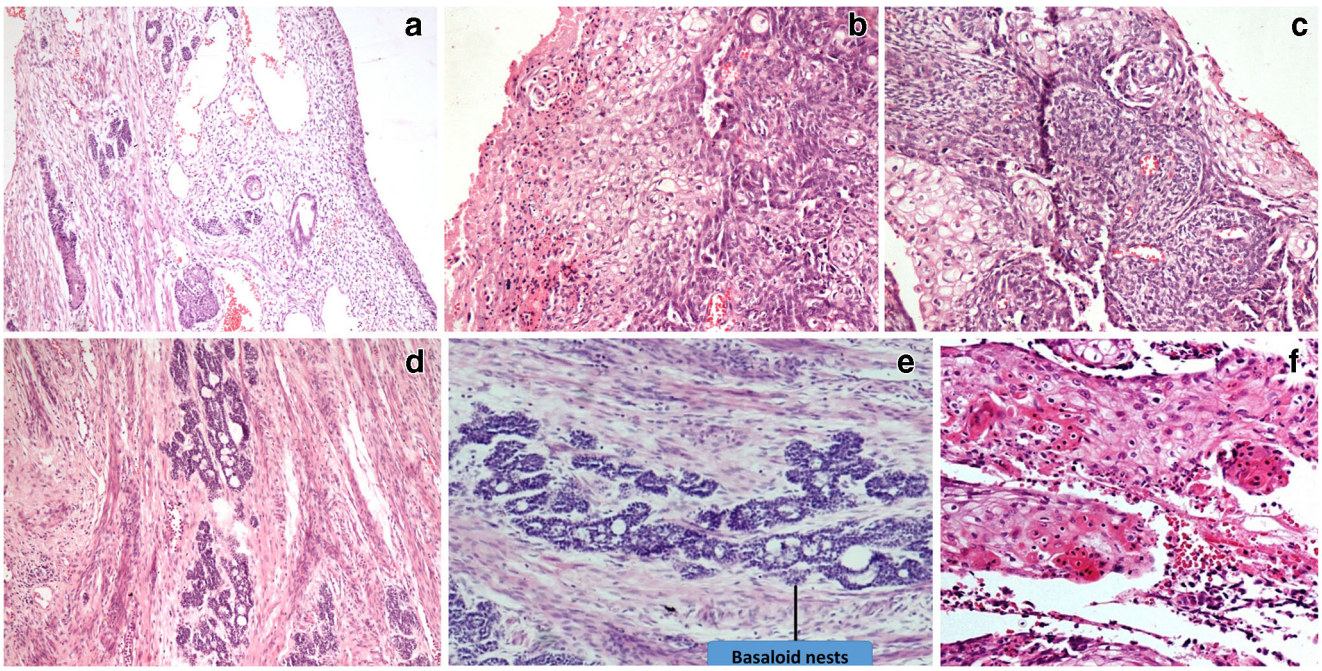

Fig. 2 Hysterectomy specimen. **a** (hematoxylin and eosin, × 10) high grade squamous intraepithelial lesion. **b** (Hematoxylin and eosin, × 20) an invasive component of squamous cell carcinoma. **c** (Hematoxylin and eosin, × 40) basal cells arising from squamous areas. **d, e** (Hematoxylin and eosin, × 40) few areas showing glandular differentiation. **f** (hematoxylin and eosin, × 40) Invasive keratinizing squamous cell carcinoma

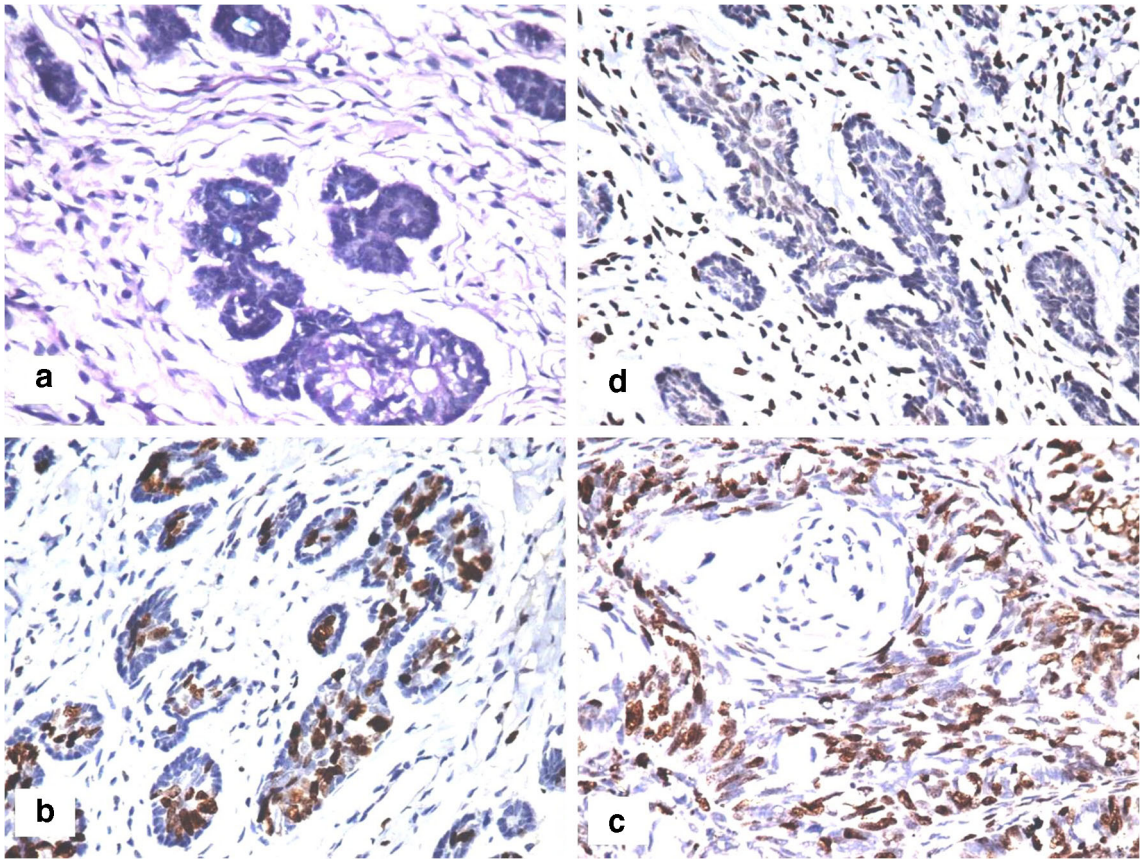

Fig. 3 Hysterectomy specimen. **a** Alcian Blue–periodic acid–Schiff stain showing Alcian Blue–positive secretions in the glandular component. Ki67 staining showed three types of staining pattern—**b** occasional glandular areas showing weak to moderate staining (15%) **c** squamous cell carcinoma component showed strong nuclear positivity with much higher labelling index (40%) and **d** most of the glandular or adenoid areas showing weak or negative staining

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

Statement of Informed Consent Written informed consent was taken from the patient with guarantee of confidentiality.

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