



Glandular Paget's Disease of the Male Nipple

Jong Hyuk Moon, Hye Soo Ko, Ji Won Byun, Gwang Seong Choi, Jeonghyun Shin

Department of Dermatology, Inha University Hospital, Inha University School of Medicine, Incheon, Korea

Dear Editor:

Paget's disease (PD) is a skin cancer characterized by epidermal infiltration of glycogen-rich vacuolated cells, termed Paget's cells, which usually present as single cells or nests^{1,2}. Otherwise, extramammary PD commonly shows the various patterns of Paget's cells including the glandular pattern. Shousha³ reported an unusual case of nipple PD where some Paget's cells showed the glandular arrangement in the epidermis. Here, we report another case of PD showing the glandular pattern.

A 68-year-old male patient presented with a 1-month history of a nonpruritic erythematous annular patch on his right areolar area (Fig. 1). Microscopic examination of a punch biopsy sample revealed atypical ductal structures with mild cellular atypia in the epidermis (Fig. 2A). Immunohistochemical results were positive for atypical ductal cells, cytokeratin 7, and carcinoembryonic antigen (CEA), and negative for cytokeratin 20 (Fig. 2B). On physical examination, no palpable lymph node or mass was detected on either breast, the axillae, or the neck. No underlying malignancy was found on computed tomography of the abdomen, pelvis, and chest and breast ultrasonography.

Mohs micrographic surgery (MMS) was performed for further histological evaluation and for treatment of the lesion. The nipple was preserved because the histological specimens of MMS showed no possible involvement and for cosmetically better result. The surgical specimen removed by MMS showed the glandular structures as well as single cells and nests of Paget's cells in the epidermis (Fig. 2C). No ductal carcinoma was found in the deep dermis. Immunohistochemistry results were as follows: CEA(+),

periodic acid-Schiff (PAS) and dPAS(+) only at the intraluminal material and S-100(-). The patient has been followed-up regularly with radiological evaluation for 3 years without recurrence.

This case was notable in that the patient showed glandular clusters of Paget's cells, along with single cells and nests in the epidermis, without underlying breast cancer. Almost all mammary PD involves underlying breast cancer. However, there have been rare reports of mammary PD without underlying malignancy¹, which are also referred to as mammary PD stage 0, or extramammary PD of the breast¹. It has been suggested that Toker cells may be potential precursor cells in cases of PD without underlying breast cancer⁴. Toker cells are clear cells of nipple epidermis⁵, with morphological and immunohistochemical characteristics similar to those of Paget's cells. To the



Fig. 1. A nonpruritic erythematous annular patch on the right areolar area.

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Corresponding author: Jeonghyun Shin, Department of Dermatology, Inha University Hospital, 27 Inhang-ro, Jung-gu, Incheon 22332, Korea. Tel: 82-32-890-2238, Fax: 82-32-890-2236, E-mail: jshin@inha.ac.kr

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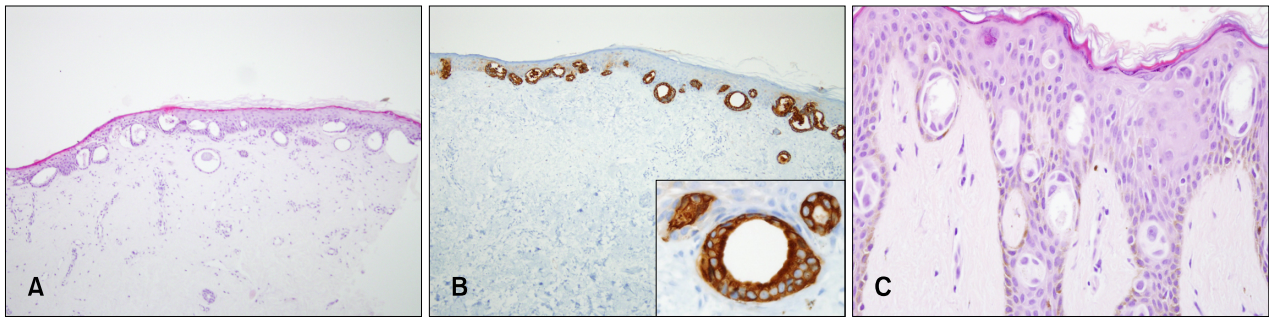


Fig. 2. (A) Punch biopsy revealed ductal structures with mild cellular atypia in the epidermis (H&E, $\times 100$). (B) Immunohistochemistry for cytokeratin 7 was positive in the ductal cells ($\times 40$; inset, $\times 400$). (C) The surgical specimen from Mohs micrographic surgery showed intraepidermal glandular structures, and Paget's cells—both single cells and nests (H&E, $\times 400$).

best of our knowledge, this is the second case of mammary PD showing the glandular pattern. We suggest that this case is one of primary PD, which might be a malignant disease of Toker cells.

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