Papillary Poroid Hidradenoma

To the Editor:

Poroid hidradenoma (PH) is a rare eccrine neoplasm, described by Abenoza and Ackerman,[1] and so-called because of its morphology intermediate between a poroma and a hidradenoma. As they are often cystic and superficially located, fine needle aspiration cytology has been useful in establishing a suspect of PH,[2,3] but the diagnosis still relies on the histological examination and the findings of the typical cell population. We describe a case of PH with prominent papillary features, arising in the thigh of a 70-year-old white man who presented with a 2-cm reddish lesion. Clinically, the lesion was tender, localized in the deep dermis, and covered by normal-looking epidermis; it was surgically removed in fragments, with sparing of the epidermis, and sent for histological examination.

On light microscopy, it presented as a cystic lesion refilled by multiple papillae (Fig. 1), sometimes with large stalks, bordered by monomorphous, cytologically bland, poroid cells (Fig. 2), and larger eosinophilic cuticular cells showing evident ductular differentiation (Fig. 2, inset). At immunohistochemistry, the former were positive with cytokeratin AE1/AE3, whereas the latter reacted with cytokeratin 7, EMA (Fig. 3A), and, focally, CEA (Fig. 3B). Neither necrosis en masse nor atypia or increased mitotic activity was noted.

Our case fulfills morphological criteria for PH, as already described,[4–9] namely, it is confined to the dermis, it has a prominent cystic component, and it is composed of a monomorphous population of either basaloid cells or large cuticular cells. Its immunohistochemical profile suggests that it is closely related to poroma,[8,9] and it is in keeping with an eccrine derivation.

A quite unusual finding is the prominent papillary architecture, which expands the spectrum of growth patterns[8]; in the presence of papillary epithelial fronds, when dealing with an adnexal skin tumor, a diagnosis of PH should therefore be considered.

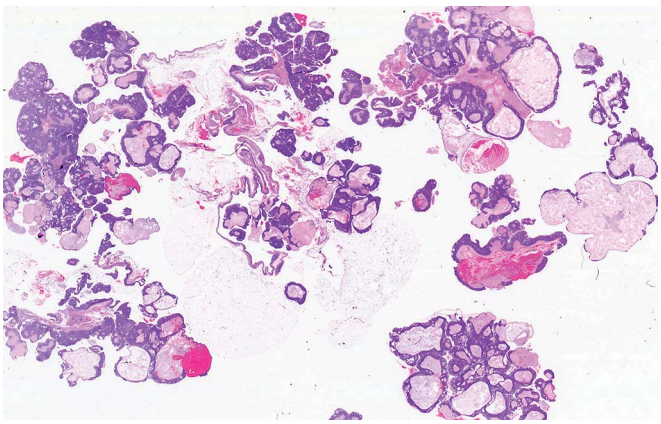

FIGURE 1. At low power, the papillary pattern of growth of the lesion is apparent.

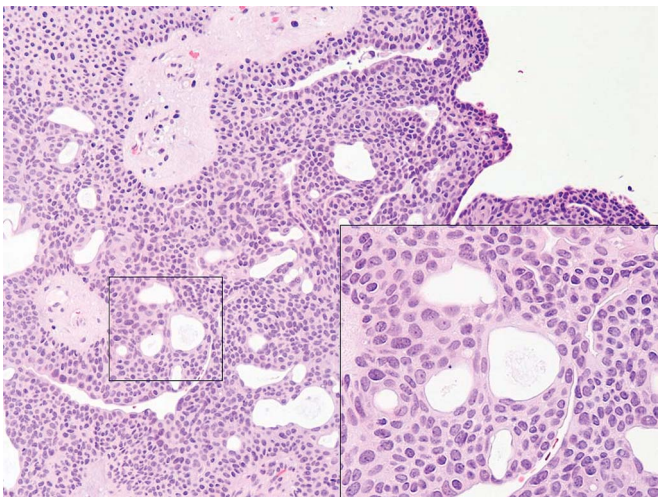

FIGURE 2. The lesion is composed of small, dark, poroid cells admixed with larger eosinophilic cuticular cells (inset).

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REFERENCES

1. Abenoza P, Ackerman AB. *Neoplasm With Eccrine Differentiation*. Philadelphia, PA: Lea & Febiger; 1990:113–185, 311–350.
2. Layfield LJ, Mooney E. Fine needle aspiration cytology of poroid hidradenoma. *Acta Cytol*. 1998;42:1064–1066.
3. Hoshida Y, Hanai J, Matsushita N, et al. Poroid hidradenoma: report of a case with cytological findings on fine needle aspiration. *Acta Cytol*. 1999;43:471–474.
4. Requena L, Sanchez M. Poroid hidradenoma, a light microscopic and immunohistochemical study. *Cutis*. 1992;50:43–46.
5. Ichioka S, Yamada A. A case of cystic dermal duct tumor corresponding to poroid hidradenoma. *J Dermatol*. 1993;20:554–557.
6. Misago N, Kohda H. A single lesion demonstrating features of eccrine poroma and poroid hidradenoma. *J Dermatol*. 1995; 22:773–779.
7. Cho S, Kim J, Shin J, et al. Poroid hidradenoma. *Int J Dermatol*. 2001;40:62–64.
8. Salem A, Abha M, Wedad H. Vulvar poroid hidradenoma. *Am J Dermatol*. 2002;24: 523–525.
9. Liu H, Chang Y, Chen C, et al. Histopathological and immunohistochemical studies of poroid hidradenoma. *Arch Dermatol Res*. 2006;297: 319–323.

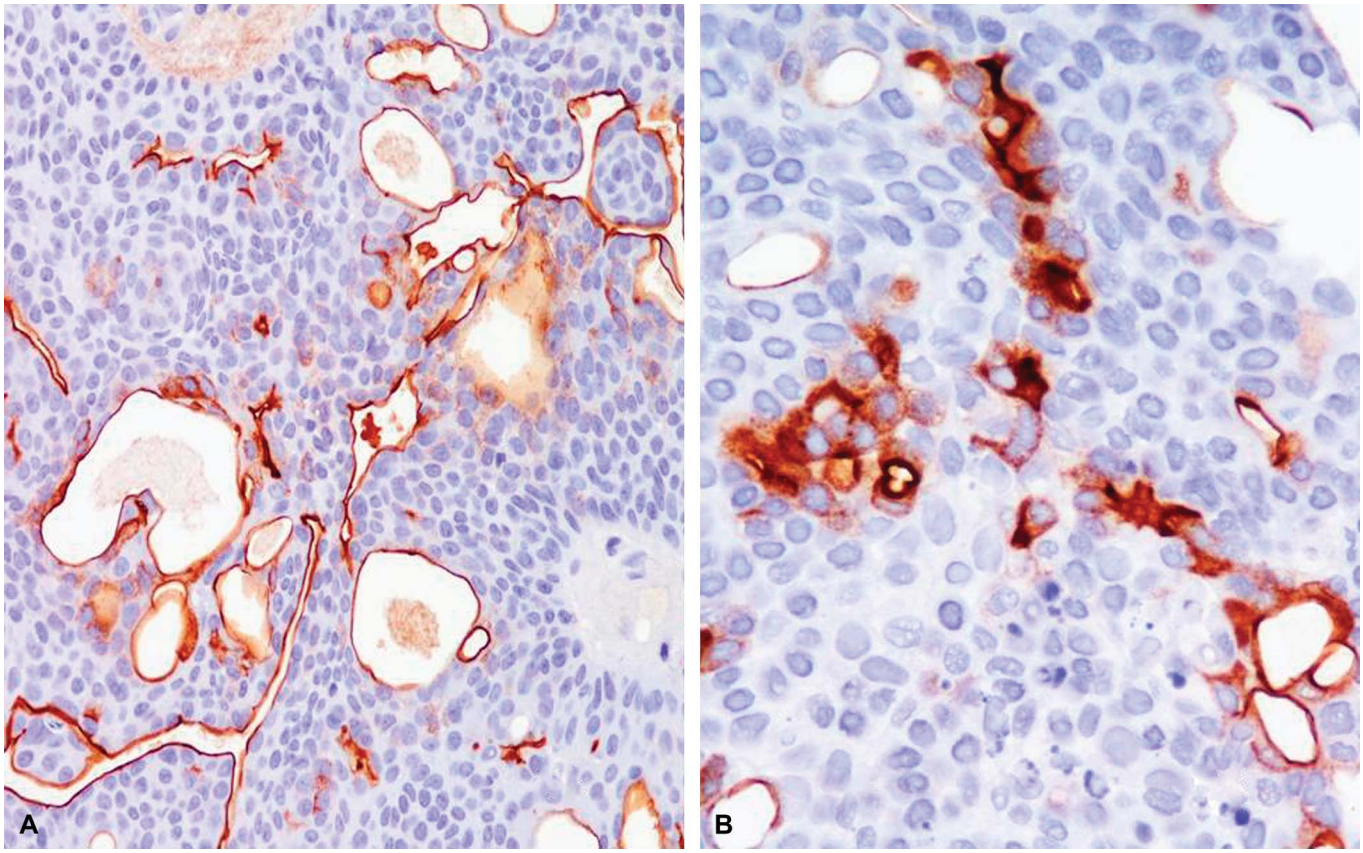

FIGURE 3. At immunohistochemistry, ductular differentiation is highlighted by epithelial membrane antigen (A) and CEA (B). CEA, carcinoembryonic antigen.

Refractory, Concomitant, Cutaneous, and Systemic Lymphomas of Discordant B-Cell and T-Cell Lineages

To the Editor:

Concomitant lymphomas of different lineages are uncommon in immunocompetent hosts. We report 2 patients with progressive systemic lymphomas and concomitant cutaneous lymphomas of discordant B- and T-cell phenotypes. Despite progressive disease in both sites, durable remission was achieved after myeloablative allogeneic hematopoietic stem cell transplantation (HSCT).

Case 1 is a 28-year-old man with stage IIIA nodular sclerosing Hodgkin lymphoma was treated with ABVD × 8 courses (adriamycin, bleomycin, vinblastine, and dacarbazine). Three years later, papular lesions appeared in the right hand and left arm. Skin biopsies showed dense lymphoid infiltrations around the adnexa and blood vessels up to the subcutis (Fig. 1A). The infiltrate consisted of $CD20^+$ B cells (Fig. 1B) and κ-restricted plasma cells, compatible with mucosa-associated lymphoid tumor (maltoma). The lesions were treated by local irradiation. Hodgkin lymphoma relapsed 3 years later and was treated with ASHAP × 3 courses (adriamycin, methylprednisolone, cytosine arabinoside, and cisplatinum) followed by autologous HSCT (conditioning: carmustine, etoposide, cytosine arabinoside, and melphalan). Lymphadenopathy recurred a year later, and an excision showed Reed–Sternberg cells, together with sheets of unrelated malignant CD3-expressing but ALK^- T cells (Figs. 1C, D). Polymerase chain reaction showed clonal T-cell receptor gene rearrangement, compatible with an unrelated peripheral T-cell lymphoma. The disease did not respond to ICE × 4 courses (ifosfamide, carboplatin, and etoposide). Concomitantly, maltoma relapsed in the left ankle. An allogeneic HSCT from a matched unrelated donor (conditioning: cyclophosphamide and total body irradiation) was performed at progressive disease. At 7-year follow-up, there was no skin or nodal lesions.

Case 2 is a 58-year-old woman presented with stage IV follicular lymphoma (Figs. 2A, B) with generalized lymphadenopathy and a lumbar epidural mass with nerve root encasement. A marrow biopsy was negative, and she was treated with CVP × 6 courses (cyclophosphamide, vincristine, and prednisolone) and radiotherapy. Five years later, she relapsed with thoracic and abdominal lymph nodes and a right thigh ulcer. A skin biopsy showed an unrelated primary cutaneous $CD30^+$ lymphoproliferative disorder (Figs. 2C, D). Both lesions

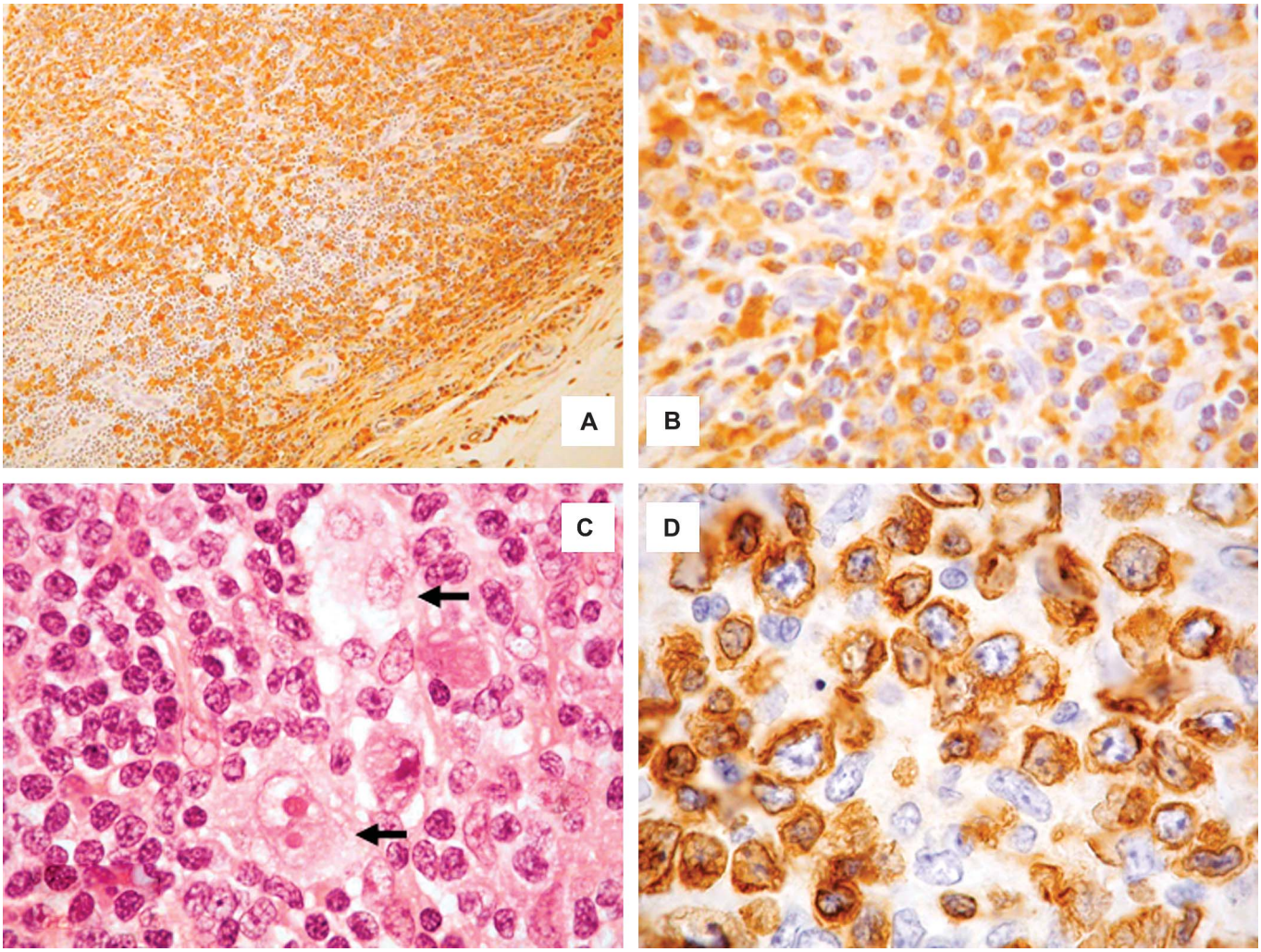

FIGURE 1. A, B, Skin biopsy showing mucosa-associated B-cell lymphoma (maltoma) consisting of dense infiltrates of $CD20^+$ B cells with prominent periadnexal infiltration (B). C, D, Lymph node biopsy showing RS cells (arrows) interspersed by sheets of T cells. The RS cells showed classical Hodgkin phenotype: $CD15^+$, $CD30^+$, $CD20^+$, $CD3^-$, and EBV-LMP1$^+$. The T cells were $CD3^+$ (D), $CD5^+$, $CD30^+$, $CD20^-$, $CD15^-$, ALK-1^-, and Epstein–Barr virus latent membrane protein-1 and showed clonal TCR rearrangement using TCRγ primers. RS, Reed–Sternberg; TCR, T-cell receptor.

persisted after Promace-Cytabom × 6 courses (cyclophosphamide, doxorubicin, etoposide, bleomycin, vincristine, methotrexate, and prednisolone) and DHAP × 3 courses (cisplatin, cytosine arabinoside, and dexamethasone). She underwent an allogeneic HSCT from a matched sibling (conditioning: fludarabine–total body irradiation) with active disease. At 1 year, there was clinical or radiological evidence of disease.

Concomitant cutaneous and systemic lymphomas with discordant B- and T-cell lineages had been reported.[1] This may reflect a publication bias because the lineage discordance obviated molecular studies to show clonal independence.[2] Etiologically, a chance occurrence is possible, especially if 1 or both lesions are of chronic relapsing nature (e.g., mycosis fungoides and chronic lymphocytic leukemia).[1,3] The second malignancy may also be attributed to the carcinogenic chemotherapy, reduced immunosurveillance, or skewed reactive lymphoproliferation, caused by the first lymphoma.[1,4] Sequential lymphoma is also a feature among immunocompromised hosts,[5] but our cases did not have human immunodeficiency virus/human T-lymphotropic virus-1 exposure, autoimmune disease, or opportunistic infections. Clinically, the refractory double (and triple) lymphoma pathology presented a challenge. Many novel antilymphoma agents are lineage specific.[6] A nonlineage-specific graft-versus-lymphoma effect proved to be a viable therapeutic option.[7] The new immune system also eradicated any theoretical immunological deficit, resulting in lymphoma propensity. Prompt and appropriate referral may therefore be prudent for oncologists and dermatologists encountering such unusual combination of lesions.

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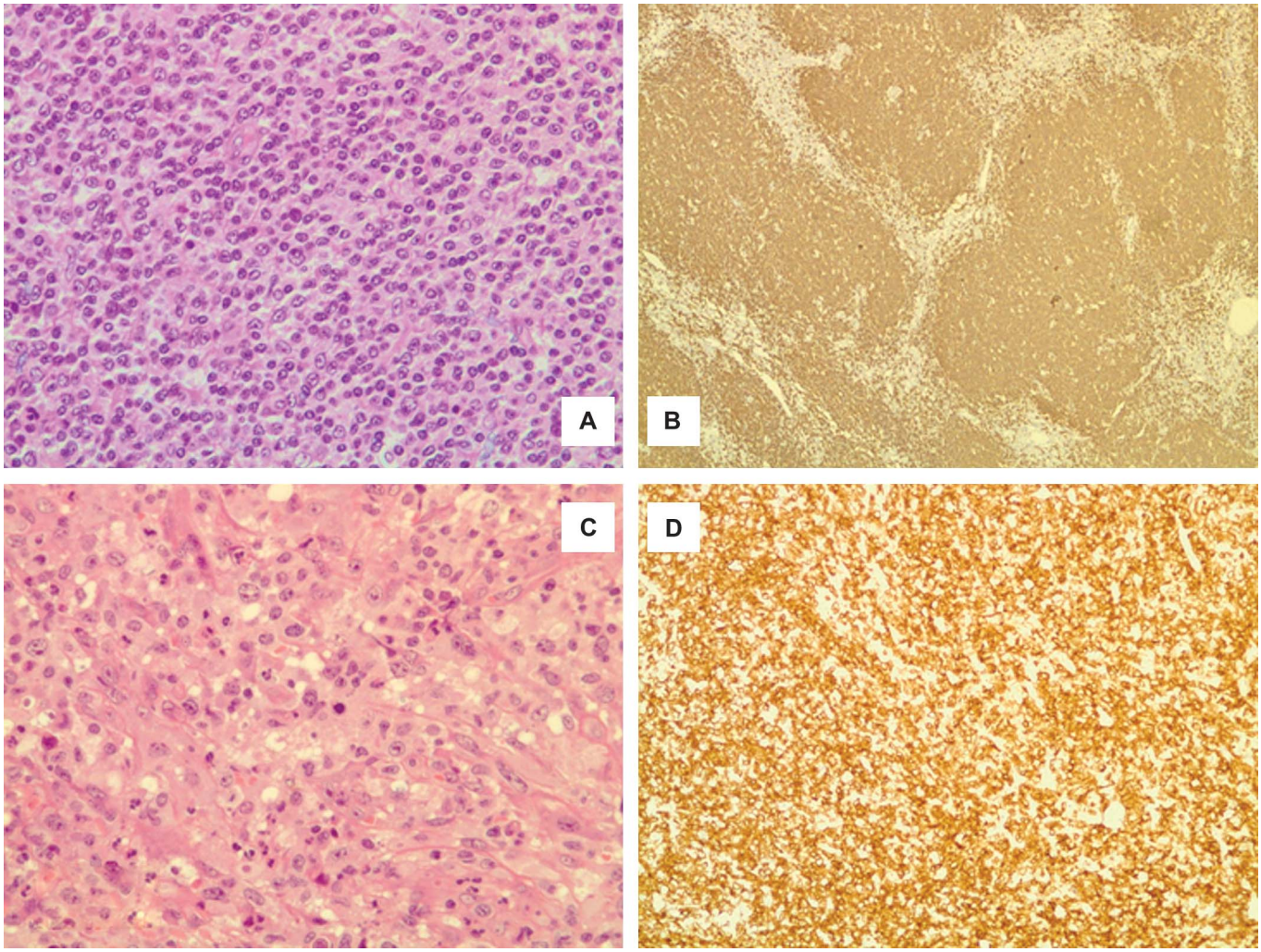

FIGURE 2. A, B, A lymph node biopsy with effaced architecture with coalescing follicles composed of small cleaved cells and occasional centroblasts (H&E, ×400). The tumor cells are positive for CD20 (B, ×40) and CD10 but were negative for CD3, CD5, and cyclin D1. Molecular study showed clonal immunoglobulin H rearrangement. C, D, Skin biopsy with dense lymphoid infiltration by medium to large malignant T cells (H&E, ×400), with cellular atypia and frequent mitosis. The tumor cells were diffusely and strongly positive for CD2 and CD30 (Fig. 1D, ×100) and patchy for CD3 with aberrant loss of CD5. Staining for CD20, CD79a, CD10, ALK-1, and CD56 were negative. Molecular study by TCRγ primers demonstrated clonal T-cell proliferation. H&E, hematoxylin and eosin; TCR, T-cell receptor.

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REFERENCES

1. Barzilai A, Trau H, David M, et al. Mycosis fungoides associated with B-cell malignancies. *Br J Dermatol*. 2006;155:379–386.
2. Robak E, Jesionek-Kupnicka D, Robak T, et al. Primary cutaneous marginal zone B-cell lymphoma in a patient with chronic lymphocytic leukaemia. *Br J Dermatol*. 2007;157:591–595.
3. Sheibani K, Forman SJ, Winberg CD, et al. Coincidence of B-cell chronic lymphocytic leukemia and cutaneous T-cell lymphoma (mycosis fungoides): immunologic characterization by monoclonal antibodies. *Blood*. 1983;62:1176–1181.
4. Martinez A, Pittaluga S, Villamor N, et al. Clonal T-cell populations and increased risk for cytotoxic T-cell lymphomas in B-CLL patients: clinicopathologic observations and molecular analysis. *Am J Surg Pathol*. 2004;28:849–858.
5. Kempf W, Margolin DH, Dezube BJ, et al. Clinicopathological characterization of an HIV-2-infected individual with two clonally unrelated primary lymphomas. *Am J Hematol*. 2000;65:302–306.
6. Bolli N, Di Ianni M, Simonetti S, et al. Treating two concur3rent B-cell and T-cell lymphoid neoplasms with alemtuzumab monotherapy. *Lancet Oncol*. 2004;5:64–65.
7. Schmitz N, Dreger P, Glass B, et al. Allogeneic transplantation in lymphoma: current status. *Haematologica*. 2007;92:1533–1548.