DPLM, our patient showed only partial response despite fifteen weeks of long term application. This might imply that the pathogenesis of two diseases is different. It is known that tacrolimus inhibits tumor necrosis factor (TNF)-α secretion in human keratinocytes and transforming growth factor (TGF)-β-nduced collagen synthesis. TNF-α and TGF-β stimulate glycosaminoglycan synthesis from skin fibroblast, and Rongioletti et al.[5] suggested that inhibiting these cytokines might be the mechanism of the tacrolimus on DPLM. The difference of the contributing proportion of these cytokines in DPLM and APPM might be the reason for different response to tacrolimus.

Although the etiology of APPM is yet unknown, our patient showed familial occurrence and it raises the possibility of the genetic role in APPM pathogenesis along with previous reports of familial occurrences[1].

Our case gives some notable points on the pathophysiology of APPM, and we hope that this case may add to the growing body of literature of APPM.

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Cutaneous Metastasis of Rhabdomyosarcoma Originated from Maxillary Sinus in a Young Adult Female

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Dear Editor:

Rhabdomyosarcoma (RMS) is a malignant mesenchymal neoplasm showing skeletal myogenic differentiation that usually arises on the head and neck, genitourinary tract, or soft tissue of the extremities[1]. Cutaneous metastases of RMS rarely reported in English literature; between 1966 and 2014 only 14 cases were reported[2]. Among them, only four cases were in adults[2].

A 21-year-old female was referred to department of dermatology for asymptomatic multiple cutaneous nodules. On physical examination, multiple, various sized, erythematous nodules were seen on the left breast (Fig. 1). In past medical history, she had 2.5-years history of RMS (embryonal subtype, clinical group II, stage III) in maxil-

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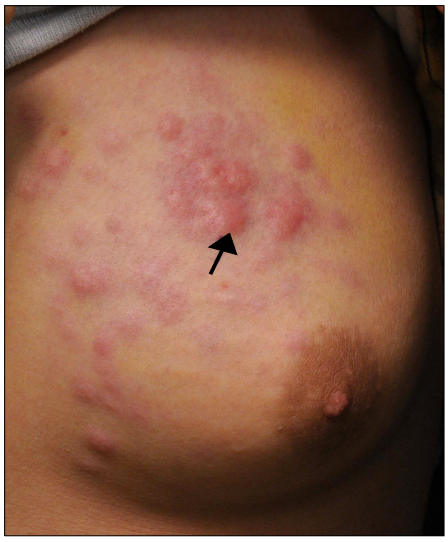

Fig. 1. Multiple, erythematous nodules on the upper and lower inner quadrants of the left breast.

lary sinus. After initial diagnosis, she received RMS-specific protocols of chemotherapy and radiotherapy for six months, and achieved complete remission. However, after 1-year, both breast metastases were detected. Accordingly, she received treatment again, but achieved only partial response.

A punch biopsy was taken from the nodule on the upper quadrant. Histopathological examination revealed dense infiltration of primitive ovoid neoplastic cells through the dermal collagen bundles (Fig. 2A). These cells had darkly stained hyperchormatic nuclei and scant cytoplasm (Fig. 2B). The neoplastic cells showed positivity for vimentin, myoglobin, desmin and myogenin, but negative signal for MyoD1 (Fig. 2C~G). Although one of the markers for skeletal muscle is negative, these findings were consistent with metastatic RMS. Then, she underwent various regimens of chemotherapy, but showed disease progression

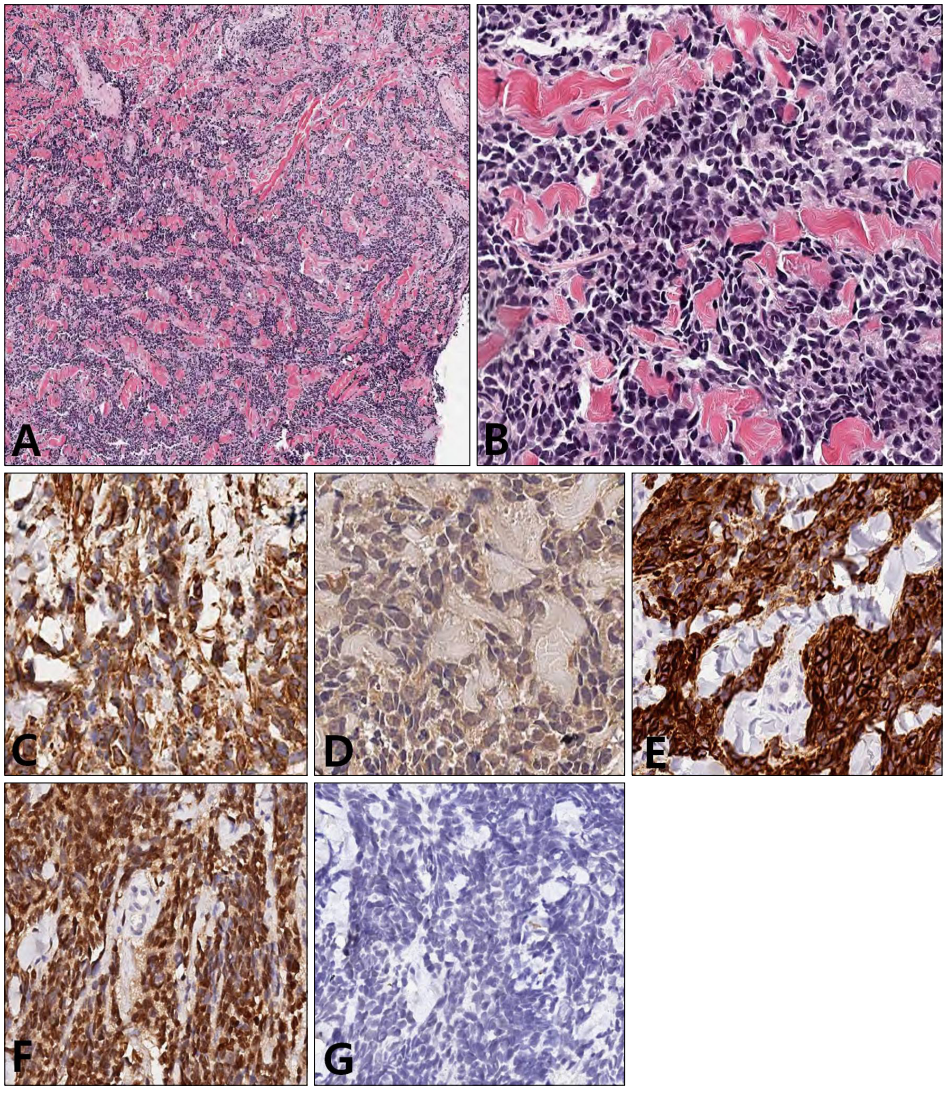

Fig. 2. Histological features of the skin nodule. (A) Dense infiltration of primitive ovoid neoplastic cells through the dermal collagen bundles (H&E, ×40). (B) Atypical cells with darkly stained hyperchormatic nuclei and scant cytoplasm (H&E, ×200). Positive immunostaining for (C) vimentin, (D) myoglobin, (E) desmin, and (F) myogenin, but negative for (G) MyoD1 (C~G: ×200).

such as multiple bone metastases. Eventually, she died after 4 years of initial diagnosis and 15 months of skin metastasis.

The soft tissue sarcomas represent <1% of all adult solid malignancy, and the RMS accounts for only 3% of all soft tissue sarcomas in adults[1]. Furthermore, the metastasis to skin and primary cutaneous involvement of the RMS are extremely rare[2-4].

It is often confusable when differentiate the cutaneous metastases of RMS from other skin neoplasms that reveal small, round blue cells[1,2]. The immunohistochemistry is helpful to make the diagnosis of RMS, since they show positivity for MyoD1, myogenin, and desmin[1,2]. However, our patient failed to show staining for MyoD1. The MyoD1 and myogenin are markers of rhabdomyoblastic differentiation and expressed in RMS except the pleomorphic subtype[5]. Especially, MyoD1 is essential for early stages of myogenesis, whereas myogenin is later[5]. In our case, the immunohistochemistry showed positivity only for myogenin. It probably implies that the block of maturation appeared at a later stage of myogenesis. However, false negativity cannot be ruled out, because the MyoD1 and myogenin have lower sensitivity than desmin due to technical reasons[5]. Therefore, Marburger et al.[3] who reported 11 cases of primary RMS, recommended second skeletal muscle specific marker may be necessary in some instances.

In conclusion, although the skin metastases are quite rare, the RMS should be included as the origin of the metastasis even in the adult. In addition, when potential diagnosis is RMS such as desmin-positive primitive tumors, both MyoD1 and myogenin are necessary to confirm the diagnosis. Finally, further evaluation is needed to establish accurate sensitivity of MyoD1 and myogenin for RMS.

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