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■ BRIEF CASE REPORT ■

Primary Leiomyosarcoma of Adrenal Gland with Tissue Eosinophilic Infiltration

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Eosinophils are associated with numerous disorders, including helminthic parasitic infections, allergic diseases, and some tumors.[1] Although eosinophils are commonly encountered in human solid tumors, their functional role in neoplasm remains a matter of controversy.[1] The correlation of tumor-associated tissue eosinophilia (TATE) with prognosis has shown variable results in several malignancies.[1-4] Primary adrenal leiomyosarcoma is a very rare malignant mesenchymal tumor with smooth muscle differentiation. Twenty-two cases have been reported in the English[5,6] and Korean[7] literature. The association of tissue eosinophilia with leiomyosarcoma is very rare, with only one case of uterine leiomyosarcoma reported in the literature.[4] We experienced a very rare case of primary adrenal leiomyosarcoma with marked tissue eosinophilia, it is the first case reported in the literature.

CASE REPORT

A 28-year-old man presented with a three-month history of right flank pain, associated with persistent and stabbing pain and weight loss. Computed tomography showed a well-circumscribed heterogeneously enhancing 13.8-cm mass located at the right suprarenal area (Fig. 1). No metastatic lesion was noted. White blood cell count and eosinophil differential count were within the normal range. Biochemical examination revealed no functional tumor of the adrenal gland. The patient had no signs or symptoms of human immunodeficiency virus or Epstein-Barr virus infection. The patient underwent a right open adrenalectomy. The resected adrenal gland mass showed a large lobulated mass (15×13×6 cm, 792 g) almost replacing the adrenal gland. The mass was a well-circumscribed and partially encapsulated solid tumor. On histologic evaluation, the spindle cell tumor showed a rim of fibrous tissue with entrapped atrophic adrenal cortical cells, geographic coagulation necrosis with surrounding fibrosis and frequent mitotic figures (25/10 high-power field) (Fig. 2A). The surgical resection margin was free of tumor. We used Fédération Nationale des Centres de Lutte Contre le Cancer (FNCLCC) or the tumor grading and the tumor grade was 3. The tumor cells were strongly cytoplasmic positive for smooth muscle actin (Fig. 3A) and desmin (Fig. 3B). However, pan-cytokeratin, CD117, S100-protein, and human melanoma black 45 were all negative. On the bases of clinical data, histomorphological features and immunohistochemical studies, we diagnosed primary adrenal leiomyosarcoma. Interestingly, there was intense tissue eosinophilia (Fig. 2B). The eosinophilic infiltration was mainly present in the viable tumor cells. The center of the necrotic area did not show either eosinophils or other inflammatory cells (Fig. 2A). A few pleomorphic tumor cells with hyperchromatic nuclei were noted (Fig. 2B). Most of the tumor cells of the tissue eosinophilia revealed vesicular nuclei, cytoplasmic shrinkage, and occasional pink round nucleoli (Fig. 2C). The patient was alive without recurrent disease on 18-month follow-up.

DISCUSSION

TATE has been observed in several cancers.[1-4] However, the

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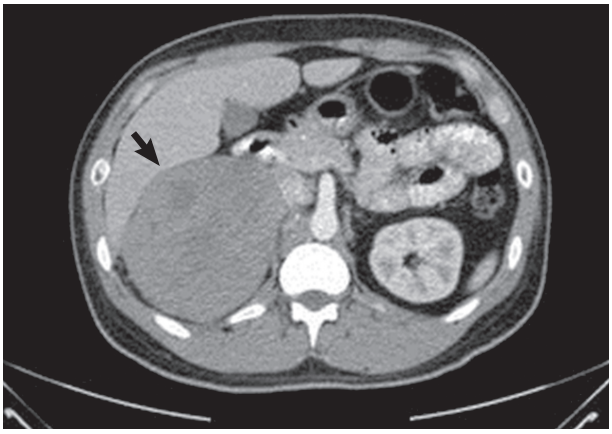

Fig. 1. Computed tomography reveals a well-circumscribed heterogeneously enhancing mass in the right suprarenal fossa, measuring 13.8×11.3×9.3 cm (arrow).

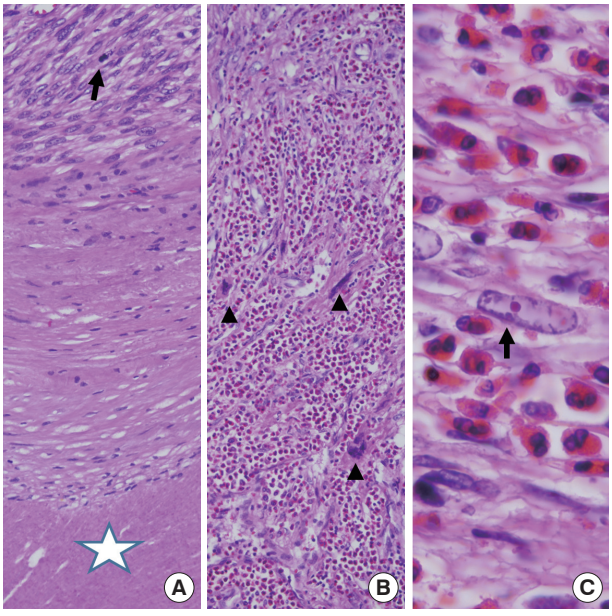

Fig. 2. Histologic features of the tumor. (A) The tumor shows geographic coagulative necrosis (star) and frequent mitotic figures (arrow). (B) There is intense eosinophilic infiltration and a few pleomorphic tumor cells (arrowheads). (C) Some nuclei show pink round nucleoli (arrow).

mechanism by which eosinophils are recruited into tumor tissue is largely unknown. It is likely that stressed or necrotic tumor cells attract and activate eosinophils by expression of molecules such as damage-associated molecular patterns. TATE seems to have tumor destructive effector functions and immunoregulative and remodeling activities.[2] Although controversy remains, TATE are associated with good prognosis.[3,4] Our case was primary adrenal leiomyosarcoma with marked tissue eosinophilia.

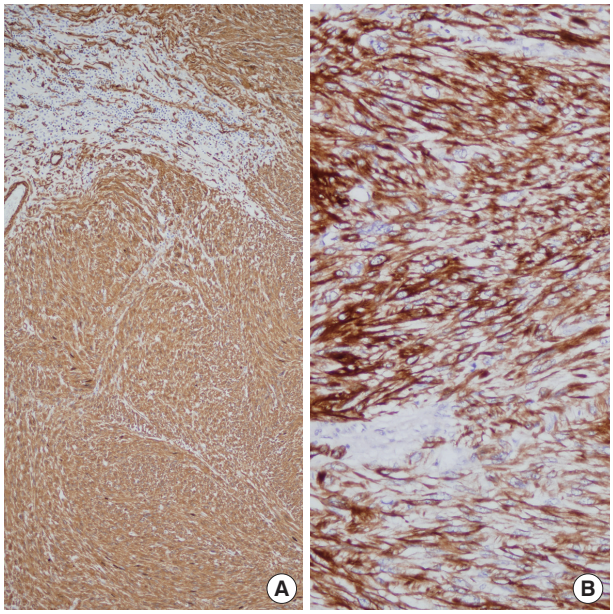

Fig. 3. The tumor cells are strongly positive for smooth muscle actin (A) and desmin (B).

Although there were a few pleomorphic tumor cells, our case was considered to be the conventional type. Based on the French grading system (FNCLCC), the sub-scores of our case were two for tumor differentiation, three for mitotic count, and one for tumor necrosis, and the histologic grade was 3. The prognosis for primary adrenal leiomyosarcoma is generally poor. Among previously reported cases, only two cases were alive without metastasis after more than 15 months follow-up.[5-7] The distinct profile of the patient is high-grade sarcoma with marked tissue eosinophilia and 18 months survival without metastasis. Although TATE was found to be a good prognostic factor in a previous report,[4] further study and longer patient follow-up are warranted to support the conclusion that TATE is a good prognostic factor for primary adrenal leiomyosarcoma.

Conflicts of Interest

No potential conflict of interest relevant to this article was reported.

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