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Myopericytoma of soft tissue (thigh)☆,☆☆,★

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Abstract The recent recognition of concept of perivascular myoid cell (PMC) phenotype and perivascular epithelioid cell (PEC) phenotype has changed the concept of angiogenic tumors, in particular "hemangioperycytoma" (HPC)-like tumors. The PMC concept has yielded myopericytoma, myofibroma, myofibromatosis, angioleiomyoma, and glomus tumors, while the PEC concept that shows melanocytic lineage has given rise to PEComa, clear cell sugar tumors, angiomyolipoma, and lymphangiomyomatosis. Here, the author reports a case of myopericytoma of soft tissue (thigh) occurring in a 47-year-old man. The patient had noticed the thigh tumor 4 years before, and was admitted. The tumor was located in subcutaneous tissue, was encapsulated, and measured 4 × 3 × 3 cm. Total excision was performed. Grossly, the tumor is a soft reddish encapsulated tumor with very well demarcation. Histologically, the tumor showed typical HPC-like features with focal nodular proliferation of HPC-like tumor cells. Immunohistochemically, the vasculatures were positive for vimentin, CD31, CD34, and factor VIII-related antigen. In contrast, the HPC-like tumor cells were positive for vimentin, α-smooth muscle antigen, h-caldesmon, bcl-2, and factor XIIIa, but negative for cytokeratins, desmin, CD31, CD34, factor VIII-related antigen, S100 protein, melanosome, CD99, and KIT. Ki-67 labeling was 15%, and p53 was mildly positive. The overall features were those of myopericytoma. The patient is now alive without recurrence 5 years after the operation. A brief literature review and differential diagnosis were made.

1. Introduction

The recent recognition of the concepts of perivascular myoid cell (PMC) phenotype and perivascular epithelioid cell (PEC) phenotype has changed the concept of angiogenic tumors, in particular "hemangioperycytoma" (HPC)-like tumors [1,2]. The PMC concept has yielded myopericytoma [3–5], myofibroma [4], myofibromatosis, angioleiomyoma, and glomus tumors, while the PEC concept that shows melanocytic lineage has led to PEComa, clear cell sugar tumor, angiomyolipoma, and lymphangiomyomatosis. These tumors belong to a spectrum of PMC and PEC tumors, and there are some overlaps among these disease entities.

HPC is a neoplasm of perivascular pericyte-like cells, and characterized by HPC-like cell proliferation with abundant vascular channels which often show staghorn-like appearances. HPC is a heterogenous tumor and it contained some specific neoplasms; the term of HPC is a waste basket diagnosis. With the recent recognition of PMC and PEC, several specific disease entities have appeared recently, as described above. Therefore, the strict HPC has become very rare.

Herein reported is the case of a 47-year-old man with myopericytoma of the thigh. A brief review of the literature

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★ Author role: The author did all except some clinical practices.
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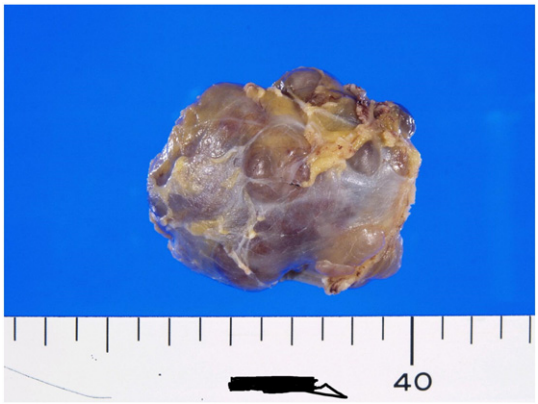

Fig. 1 Gross features of the tumor. It is an encapsulated, well defined, and reddish tumor measuring 4 × 3 × 3 cm.

and differential diagnosis were made. To the best of the author's knowledge, reports of soft tissue MPC is relatively scant; 73 cases of MPC have been reported in 15 papers.

2. Case report

A 47-year-old man noticed the thigh tumor 4 years ago, and was admitted to our hospital. The tumor was located in the subcutaneous tissue and measured 4 × 3 × 3 cm. A small biopsy revealed HPC-like tumor. A total excision was performed. Grossly, the tumor was a soft reddish tumor with encapsulation (Fig. 1). The tumor was well defined from the surrounding tissues and the excisional procedure was easy. Histologically, the tumor showed typical HPC-like features (Fig. 2A and B) with focal nodular proliferation of HPC-like tumor cells. The tumor is abundant in vasculatures (Fig. 2), and the vasculatures often showed staghorn appearances (Fig. 2A and B). The tumor is relatively heterogenous, and hypervascular and vascular-rich areas were recognized. The tumor cells had vesicular nuclei, and neither nuclear atypia nor necrosis was recognized. Secondary degeneration was recognized in a few areas. Mitotic figures were present in 5 per 50 high power fields.

An immunohistochemical study was performed with the use of Dako Envision method (Dako, Glostrup, Denmark), as described previously. The antibodies used were as follows: cytokeratin (AE 1/3, Dako), cytokeratin (CAM5.2, Beckton-Dickinson, CA, USA), CD34 (NU-3A1, Dako), S100 protein (polyclonal, Dako), desmin (D33, Dako), α-smooth muscle antigen (1A4, Dako), myoglobin (polyclonal Dako), vimentin (Vim 3B4, Dako), CD31 (JC70A, Dako), p53 protein (DO7, Dako), Ki-67 (MIB-I, Dako), melanosome (HMB 45, DAKO), factor VIII-related antigen (36B11, Novocastra, Newcastle upon type, UK), bcl-2 (124, Dako), KIT (polyclonal, Dako), CD99 (12E7, Dako), factor XIIIa (E980.1, Novocastra), and h-caldesmon (h-CD, Dako).

Immunohistochemically, the vasculatures were positive for vimentin, CD31, CD34, and factor VIII-related antigen (Fig. 3A), but negative for cytokeratins and other antigens examined. In contrast, the HPC-like tumor cells were positive for vimentin, α-smooth muscle antigen (Fig. 3B), h-caldesmon, bcl-2 (Fig. 3C), and factor XIIIa (Fig. 3D), and negative for cytokeratins, desmin, CD31, CD34, factor VIII-related antigen, S100 protein, melanosome, myoglobin, CD99, and KIT. Ki-67 labeling index was 15%, and p53 was mildly positive.

The overall features were those of myopericytoma. The patient is now alive without recurrence 5 years after the operation.

3. Discussion

The histology and immunohistochemical findings of the present study indicate that the present case is HPC-related tumor. Because the tumor cells were positive for α-smooth muscle actin and h-caldesmon, the tumor belongs to PMC tumor group which includes myopericytoma [3–5], myofibroma [4], myofibromatosis, angioleiomyoma, and glomus tumors. Of these, the present case was myopericytoma. Because the present case was negative for S100 protein and melanosome, the present tumor does not belong to the PEC tumor group which includes PEComa, clear cell sugar tumor, angiomyolipoma, and lymphangiomyomatosis [1]. The

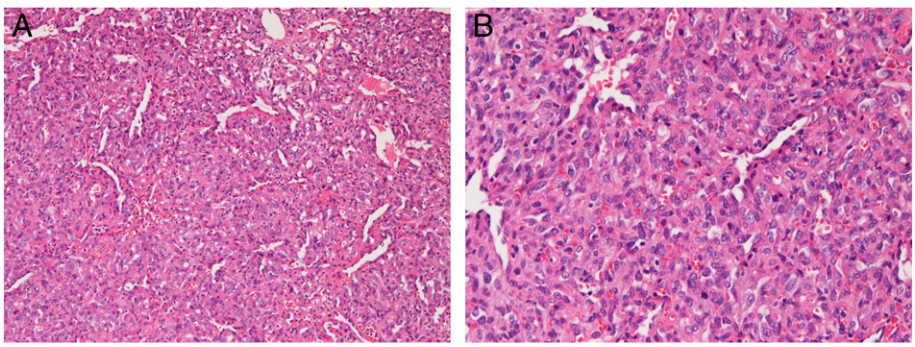

Fig. 2 Histological features of the tumor. A: Low power view of the tumor. The tumor shows hemangiopericytomatous pattern. HE, ×100. B: The tumor consists of fine vasculatures and perivascular tumor cells, creating hemangiopericytomatous pattern. The vasculatures show staghorn appearances. The tumor cells are free of atypia. Vague nodular pattern is seen around the vasculatures. HE, ×200.