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A case of atypical benign fibrous histiocytoma

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A case of atypical benign fibrous histiocytoma is reported. A 62-year-old Japanese female visited our clinic because of an asymptomatic solitary lesion on the skin of the left leg. Physical examination revealed a polypoid mass lesion (2.5×2.3×1.8 cm) with central erosion. The lesion began with a 1 mm- sized papule and slowly enlarged over the 20 years. Clinical diagnosis was a malignant tumor such as dermatofibrosarcoma protuberans, atypical fibroxanthoma or adnexal tumors. Biopsy of the polypoid lesion was carried out.
Histopathological examination revealed a polypoid lesion consisting of proliferation of fibroblast-like spindle cells in the dermis. Large atypical cells with pleomorphic nuclei were occasionally observed but mitotic figures were rare. From immunohistochemical results (CD68, Factor-XIII, MIB-1 labeling index), we diagnosed this case as "atypical benign fibrous histiocytoma (ABFH)".
Clear distinction has not been made between ABFH, a variant of benign fibrous histiocytoma, and atypical fibroxanthoma, which is a variant of malignant fibrous histiocytoma. Here we report a case of ABFH with a diagnosis of the neoplasm.

Key words : Atypical benign fibrous histiocytoma, benign fibrous histiocytoma, atypical fibroxanthoma, malignant fibrous histiocytoma, MIB-1 labeling index

INTRODUCTION

Atypical benign fibrous histiocytoma (ABFH) is a variant of benign fibrous histiocytoma characterized by histologic atypism and is also called dermatofibroma [1], cutaneous histiocytoma [2], atypical cutaneous fibrous histiocytoma [3], or dermatofibroma with monster cells [4]. We report here a case of ABFH.

CASE REPORT

A 62-year-old Japanese woman visited our hospital on October 23, 2002 because of a nodule on the left leg. The lesion began as a solitary and asymptomatic red papule 1 mm in diameter in 1981 and enlarged slowly. The patient had no particular medical or family history. On the physical examination at the initial visit, the lesion was an elastic-hard polypoid mass 2.5 × 2.3 × 1.8 cm in size with central erosion (Fig. 1a and 1b). Differential diagnosis included dermatofibrosarcoma protuberans, atypical fibroxanthoma and adnexal tumors. Excisional biopsy of the mass with a 2-mm lateral tumor-free margin was performed under local anesthesia.

Histopathological examination revealed a well-demarcated mass containing eosinophilic hyaline and polygonal atypical cells in the dermis, with epidermal hyperplasia and a Grenz zone (Fig. 2a). There were large atypical foam cells in the hyaline (Fig. 2b). Few mitotic figures were noted. With the impression of a malignant tumor, indirect immunoperoxidase method was carried out for further evaluation. The results showed that the bizarre cells were positive for CD68 and Factor XIII but negative for CD34, S-100, HMB-45, and Factor VIII. The MIB-1 labeling index was less than 1 % (Table 1).

On the basis of the clinical and histologic

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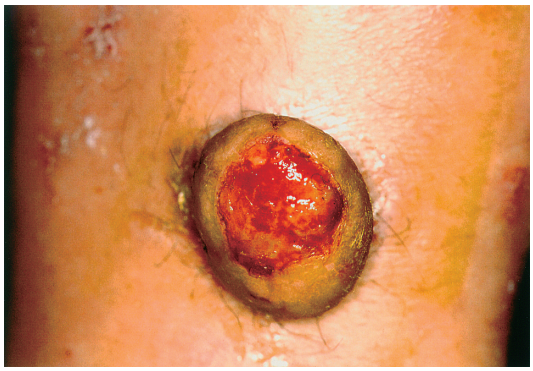

Fig. 1a Climical appearance

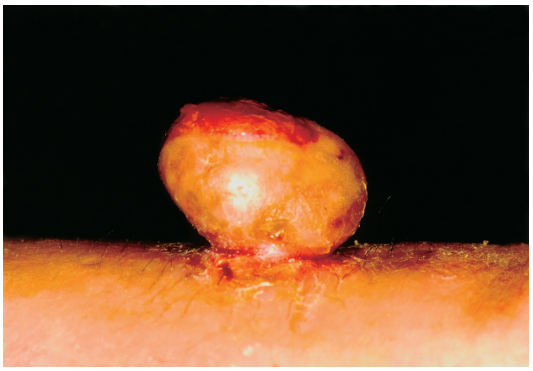

Fig. 1b Climical appearance

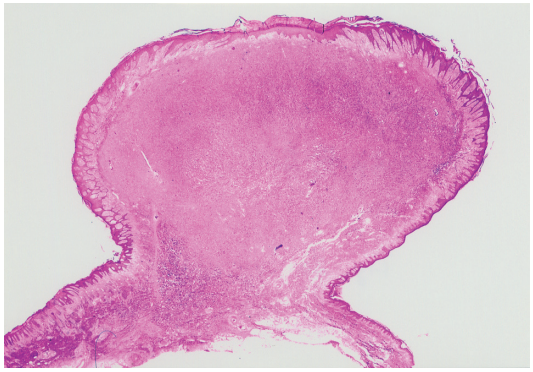

Fig. 2a Histological findings (H-E stain×1.25)

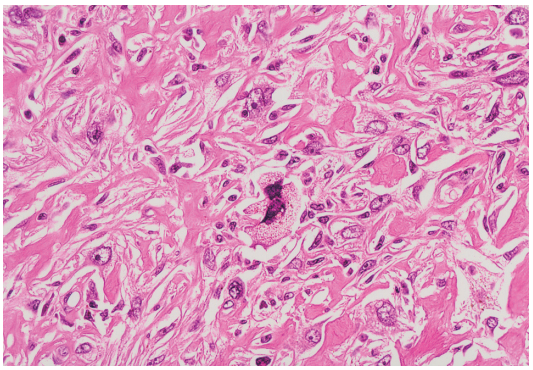

Fig. 2b Histological findings (H-E stain×100)

Table 1 Used antibodies in this case

	Companies	Dilution	Results
CD68	DAKO	×100	Positive
Factor XIII	Behring	×50	Positive
CD34	Novocastra Laboratories	×10	Negative
S-100	DAKO	×200	Negative
HMB-45	DAKO	×20	Negative
Factor VIII	DAKO	×200	Negative
MIB-1	IMMUNOTECH S.A.	×50	Labeling index: <1 %

Table 2 Comparison of Atypical benign fibrous histiocytoma and Atypical fibroxanthoma

	Atypical benign fibrous histiocytoma	Atypical fibroxanthoma
Age	middle age (average, 39 years old)	advanced age (average, 67 years old)
Location	Trunk and limbs	Head and neck
Epidermis	Acanthotic	Atrophic or flat
Grenz zone	+	−
Mitoses	− or rare	+
CD34 staining	Variable	Variable
Factor XIII staining	Variable	+
MIB-1 labeling index	Low (average, 3.3 %)	High (average, 12.2 %)
Metastasis	−	Rare

findings, we diagnosed the case as ABFH. No recurrent tumor was found for 1 year since the excisional biopsy.

DISCUSSION

The definition of ABFH is variable. Elder *et al*. categorized atypical cutaneous histiocytoma [2], atypical cutaneous fibrous histiocytoma [3], and dermatofibroma with monster cells [4] as ABFH in 1997 [1]. According to their report, the diameter of ABFH is usually 1.2 cm or less but may exceed 2.5 cm. The tumor consists of fibroblast-like spindle cells with large atypical foam cells. ABFH is regarded as a variant of benign fibrous histiocytoma (dermatofibroma). Differential diagnosis includes atypical fibroxanthoma, which is a subtype of malignant fibrous histiocytoma of the skin with paradoxically benign clinical behaviors. In contrast, Fretzin *et al*. defined ABFH as a subtype of atypical fibroxanthoma [5]. ABFH tends to occur on the trunk or limbs in young adults and does not metastasize. Histologically, the epidermis is normal or hyperplastic with a Grenz zone.

Clinical and histological aspects of ABFH are compared with atypical fibroxanthoma in Table 2 [1, 3, 4, 6 and 7]. ABFH can be differentiated from atypical fibroxanthoma, which typically has a good prognosis but may metastasize, by the absence or minimal atypia of proliferating fibroblast-like spindle cells, rare mitotic figures and cellular arrangement around individual collagen bundles although atypical cells with pleomorphic nuclei are sometimes seen. In our case, the tumor appeared on the lower limb. Histologic examination revealed a well-demarcated mass with rare mitotic figures, epidermal hyperplasia, and a Grenz zone. Hyaline, however, contained large atypical foam cells. These clinical and histologic features were not sufficient to make a diagnosis in our case. Immunostaining showed that the tumor cells were negative for CD34 but positive for Factor XIII. The MIB-1 labeling index was less than 1 %, which indicates low proliferative activity [7]. Since the tumor had histological findings characteristic of benign fibrous histiocytoma and contained atypical cells with no proliferative activity, we diagnosed the case as ABFH.

To establish the diagnostic procedures and treatment of ABFH, this case would serve as a good example for differential diagnosis. Further case collection as well as histologic and genetic studies are needed.

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