Int J Clin Exp Pathol 2013;6(9):1890-1893
www.ijcep.com /ISSN:1936-2625/IJCEP1307036

Original Article

Pigmented pilomatricoma: an underrecognized variant

Mitsuaki Ishida, Hidetoshi Okabe

Department of Clinical Laboratory Medicine and Division of Diagnostic Pathology, Shiga University of Medical Science, Shiga, Japan

Received July 21, 2013; Accepted August 16, 2013; Epub August 15, 2013; Published September 1, 2013

Abstract: The presence of melanin pigment and/or melanocytes in pilomatricoma has been rarely documented. In this study, we analyzed the incidence and clinicopathological features of pigmented pilomatricoma. Fifty-seven consecutive pilomatricoma cases from 53 Japanese patients were examined in this study. In fourteen cases (24.6%), pigmentation was observed in pilomatricoma. This variant equally affected in males and females, and the common locations were the upper arm and face. Proliferation of dendritic melanocytes was observed within basaloid cell nests in all cases, and melanin pigment was also present within the cytoplasm of the basaloid cells in 11 cases. Melanin pigment was also present in the shadow cells in 7 cases. The incidence of pigmented pilomatricoma as documented in previous reports is approximately 10%. However, our analysis revealed that pigmented pilomatricoma was found in 24.6% of Japanese cases of pilomatricoma, thus, this variant is not uncommon and may be under-recognized.

Keywords: Pilomatricoma, pigmented, melanocytes

Introduction

Pilomatricoma is a relatively common benign cutaneous adnexal tumor which is thought to have differentiated towards hair matrix and the inner sheath of normal hair follicle as well as hair cortex [1]. Pigmentation has been reported in various types of non-melanocytic cutaneous tumors, such as basal cell carcinoma, Bowen's disease, poroma, hidroacanthoma simplex, and Paget's disease [2-5]. However, the presence of melanin pigment and/or melanocytes in pilomatricoma, which is referred to as pigmented pilomatricoma, has been rarely documented [6-10]. In this report, we analyze the incidence and clinicopathological features of pigmented pilomatricoma.

Materials and methods

Case selection

The pilomatricoma cases in this study were comprised of 57 consecutive operative cases from our hospital collected between 2007 and 2012. The diagnosis was confirmed by two diagnostic pathologists independently, and the diagnostic criteria for pilomatricoma were based on the description by the World Health Organization Classification [1].

This study included 57 cases from 53 Japanese patients: one patient had three lesions and two patients had two lesions. The median age of the patients was 28.8 years (range from 0 to 77 years). The patients showed a slight female predominance (male/female 23/30). The locations of the lesions were as follows: arm (25 cases), face (15 cases), neck (6 cases), thigh (5 cases), back (3 cases), scalp (2 cases), and thoracic region (1 case).

Immunohistochemistry

Immunohistochemical stainings were performed using an autostainer (Benchmark XT system, Ventana Medical System, Tucson, AZ, USA) by the same method as previously reported [11-13]. The following primary antibodies were used: a mouse monoclonal antibody against HMB-45 (HMB-45, Novocastra Laboratories, Ltd., Newcastle upon Tyne, UK), a mouse monoclonal antibody against Melan-A (A103, Novocastra), and a rabbit polyclonal antibody against S-100 protein (Nichirei Bioscience, Tokyo, Japan).

Table 1. Clinicopathological features of pigmented pilomatricoma

Case No.	Age	Gender	Location	Melanocytic hyerplasia	Melanin pigment in the basaloid cells	Melanin pigment in the shadow cells
1	3	Female	Face	+	+	-
2	45	Female	Face	+	+	+
3	22	Female	Upper arm	+	+	-
4	14	Female	Back	+	+	-
5	43	Male	Upper arm	+	-	-
6	70	Male	Thigh	+	+	+
7	8	Female	Face	+	-	-
8	8	Female	Upper arm	+	+	+
9	16	Female	Upper arm	+	+	+
10	10	Male	Face	+	+	+
11	8	Male	Face	+	+	-
12	18	Male	Upper arm	+	+	+
13	8	Male	Scalp	+	-	-
14	14	Male	Upper arm	+	+	+

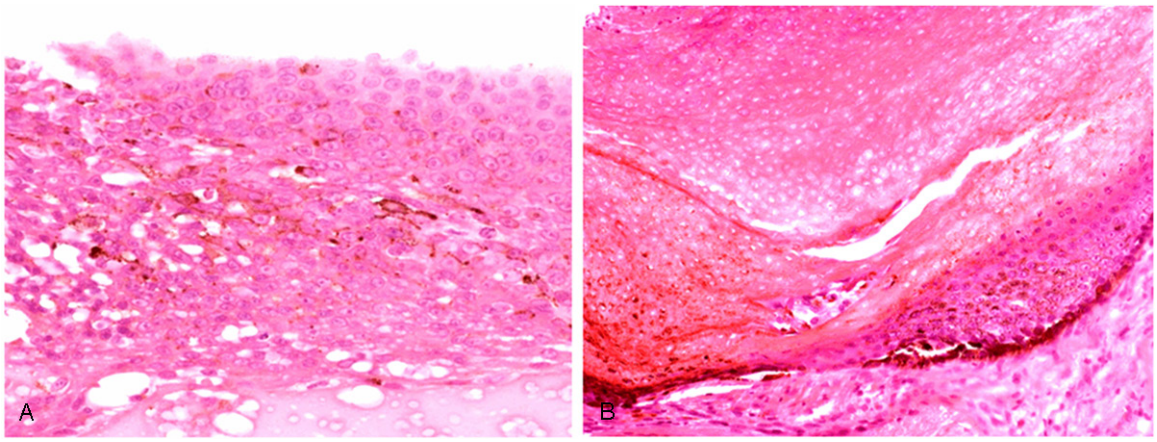

Figure 1. Histopathological features of pigmented pilomatricoma. A: Dendritic melanocytes are present within the basaloid cells, HE, x 400. B: Melanin pigment is present within the cytoplasm of the basaloid cells and shadow cells, HE, x 200.

Results

Histopathological characteristics

The characteristic histopathological features of pilomatricoma include occurrence of a relatively well-circumscribed dermal or dermal to subcutaneous nodular lesion, characterized by the presence of aggregates of basaloid cells (matrical and supramatrical cells) and filled centrally with masses of eosinophilic cornified material (faulty hair matrix) containing shadow cells. The ratio of the basaloid and shadow cells was variable, and in some cases, the shadow cells were predominant whereas the basaloid cells were scant or absent (13 cases in this series contained no basaloid cells).

Seven cases (5 cases from the upper arm and 2 cases from the thigh) had ossification within the lesion. Moreover, three cases showed a connection to the hair infundibulum.

In fourteen of 57 cases (24.6%), pigmentation was observed in the pilomatricoma. **Table 1** summarizes the clinicopathological features of the 14 cases of pigmented pilomatricoma. This variant of pilomatricoma equally affected males and females (male/female 7/7), and the average age was 20.5 years (range from 3 to 70 years). The common locations were upper arm (6 cases) and face (5 cases), and this tumor also occurred in the scalp, back, and thigh. Proliferation of dendritic melanocytes without

Table 2. The prevalence and characteristics of pigmented pilomatricoma

Case series	Prevalence (%)	Pigmented pilomatricoma/all analyzed cases	Melanocytic hyperplasia	Reference
Present study	24.6	14/57	All 14 cases containing melanocytes in the basaloid cells	
Tallon and Cerroni	10.7	3/28	All 3 cases containing melanocytes in the basaloid cells	[6]
Forbis and Helwing	7.9	19/240	3 cases containing melanocytes in the basaloid cells	[7]
Cazers et al.	8.1	3/37	1 case containing melanocytes in the basaloid cells	[8]

nuclear atypia was observed within the basaloid cell nests in all 14 cases, and melanin pigment was also present within the cytoplasm of the basaloid cells in 11 cases (**Figure 1A**) (**Table 1**). No mitotic figures were noted in the melanocytes. Melanin pigment was also present in the shadow cells in 7 cases (**Figure 1B**) (**Table 1**), however, dendritic melanocytes were not present within the shadow cells.

Immunohistochemical characteristics

The dendritic melanocytes were positive for S-100 protein and Melan-A in all cases, and some of them were also positive for HMB-45.

Discussion

In this report, we analyzed the incidence and clinicopathological features of pigmented pilomatricoma. In the present series, pigmented pilomatricoma was found in 24.6% of Japanese cases of pilomatricoma. Our analysis revealed that this variant is not uncommon and may be under-recognized. **Table 2** summarizes the previously reported series of pigmented pilomatricoma as well as the present one. Recently, Tallon and Cerroni reported 3 cases of pigmented pilomatricoma [6]. They analyzed 28 cases of pilomatricoma including a consultation case, and the prevalence rate of pigmented pilomatricoma in their series was 10.7% [6]. The incidence of pigmented pilomatricoma in the other previous reports was less than 10% (**Table 2**) [7, 8]. The reason for the difference in the incidence of pigmented pilomatricoma between the present study and the previously reported series is unknown. Although the information regarding the ethnicity of the patients with pigmented pilomatricoma in the previous reports is not available, all reports were from Europe and United States of America [6-8]. In the present case, all patients were Japanese. Therefore, this difference may be related to the ethnicity of the patients.

In the present series, melanocytic hyperplasia was observed in all cases, and melanin pigment was present within the cytoplasm of the basaloid cells in 11 of 14 cases. Pigmented pilomatricoma as reported by Tallon and Cerroni showed melanocytic hyperplasia in all 3 cases, which was also observed in the present series, and melanin pigment within the basaloid cells was observed in one of these 3 cases [6]. However, according to the other previous reports, three of 19 cases and one of 3 cases of pilomatricoma contained melanocytic hyperplasia in the basaloid cells (**Table 2**) [7, 8]. These results suggest that pigmented pilomatricoma shows both melanocytic hyperplasia and/or melanin pigments within the basaloid cells.

Pigmented pilomatricoma affects mainly young people, especially those less than 20-years-old. However, this disease can also occur in the elderly; 70-year-old and 74-year-old males were affected in the present series and in the report by Tallon and Cerroni [6], respectively. Moreover, the common sites of pigmented pilomatricoma are the face and upper arm, which are the same as conventional pilomatricoma [1].

Normal hair contains melanocytes within the hair matrix and is postulated to descend during anagen from the outer root sheath [14]. Therefore, it is not surprising that dendritic melanocytes may be present in the matrical cells of pilomatricoma [6]. Moreover, melanin pigment in the shadow cells is not an unexpected finding because normal cortical cells incorporate melanosomes. Some malignant tumor cells, such as breast cancer and squamous cell

carcinoma of the oral mucosa, have been shown to produce melanocyte chemotactic factors, resulting in melanocytic colonization within the tumor [15, 16]. Therefore, it is hypothesized that the basaloid cells can produce melanocytic trophic factors which can lead to melanocytic hyperplasia in pilomatricoma [6].

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Mitsuaki Ishida, Department of Clinical Laboratory Medicine and Division of Diagnostic Pathology, Shiga University of Medical Science, Tsukinowa-cho, Seta, Otsu, Shiga, 520-2192, Japan. Tel: +81-77-548-2603; Fax: +81-77-548-2407; E-mail: mitsuaki@belle.shiga-med.ac.jp

References

[1] Hurt MA, Kaddu S, Kutzner H, Cribier B, Schulz T, Hartschuh W. Benign tumours with follicular differentiation. In: LeBoit PE, Burg G, Weedon D, Sarasain A, editors. World Health Organization Classification of Tumours, Pathology and Genetics of Skin Tumours. Lyon: IARC Press, 2006; pp: 152-159.

[2] Lao LM, Kumakiri M, Kiyohara T, Kuwahara H, Ueda K. Sub-populations of melanocytes in pigmented basal cell carcinoma: a quantitative, ultrastructural investigation. J Cutan Pathol 2001; 28: 34-43.

[3] Ishida M, Hotta M, Kushima R, Okabe H. A case of porocarcinoma arising in pigmented hidroacanthoma simplex with multiple lymph node, liver and bone metastases. J Cutan Pathol 2011; 38: 227-231.

[4] Ishida M, Okabe H. Pigmented anal gland adenocarcinoma with associated pagetoid spread. J Cutan Pathol 2013; 40: 428-430.

[5] Ishida M, Yoshida K, Kagotani A, Arita N, Iwamoto N, Iwai M, Okabe H. Pigmented Paget's disease: A diagnostic pitfall. Diagn Cytopathol 2013; doi: 10.1002/dc.22977. [Epub ahead of print].

[6] Tallon B, Cerroni L. Where pigmented pilomatricoma and melanocytic matricoma collide. Am J Dermatopathol 2010; 32: 769-773.

[7] Forbis R Jr, Helwing EB. Pilomatricoma (calcifying epithelioma). Arch Dermatol 1961; 83: 606-618.

[8] Cazers JS, Okun MR, Pearson SH. Pigmented calcifying epithelioma. Review and presentation of a case with unusual features. Arch Dermatol 1974; 110: 773-774.

[9] Zaim MT. Pilomatricoma with melanocytic hyperplasia: an uncommon occurrence and a diagnostic pitfall. Arch Dermatol 1987; 123: 865-866.

[10] Spitz D, Fisher D, Friedman RJ, Kopf AW. Pigmented pilomatricoma. A clinical simulator of malignant melanoma. J Dermatol Surg Oncol 1981; 7: 903-906.

[11] Ishida M, Mori T, Umeda T, Kawai Y, Kubota Y, Abe H, Iwai M, Yoshida K, Kagotani A, Tani T, Okabe H. Pleomorphic lobular carcinoma in a male breast: case report with review of the literature. Int J Clin Exp Pathol 2013; 6: 1441-1444.

[12] Ishida M, Iwai M, Yoshida K, Kagotani A, Okabe H. Sarcomatoid carcinoma with small cell carcinoma component of the urinary bladder: a case report with review of the literature. Int J Clin Exp Pathol 2013; 6: 1671-6.

[13] Nakanishi R, Ishida M, Hodohara K, Yoshida T, Yoshii M, Okuno H, Horinouchi A, Iwai M, Yoshida K, Kagotani A, Okabe H. Prominent gelatinous bone marrow transformation presenting prior to myelodysplastic syndrome: a case report with review of the literature. Int J Clin Exp Pathol 2013; 6: 1677-82.

[14] Slominski A, Wortsman J, Plonka PM, Schallreuter KU, Paus R, Tobin DJ. Hair follicle pigmentation. J Invest Dermatol 2005; 124: 13-21.

[15] Konomi K, Imayama S, Nagae S, Terasaka R, Chijiiwa K, Yashima Y. Melanocyte chemotactic factor produced by skin metastases of a breast carcinoma. J Surg Oncol 1992; 50: 62-66.

[16] Satomura K, Tokuyama R, Yamasaki Y, Yuasa T, Tatehara S, Ishimaru N, Hayashi Y, Nagayama M. Possible involvement of stem cell factor and endothelin-1 in the emergence of pigmented squamous cell carcinoma in oral mucosa. J Oral Pathol Med 2007; 36: 621-624.