A Case of Trichoblastoma

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Trichoblastoma, one of the trichogenic tumors, is a rare, benign, adnexal neoplasm composed predominantly of follicular germinative cells. Clinically, it presents as a solitary, nonulcerated, skin-colored to brown or bluish-black papule or nodule situated mostly on the head and neck, particularly on the scalp and face of adults. Analysis of the distribution of specific cytokeratins (CKs) can provide a better understanding of the morphologic and/or growth features, histogenesis and differentiation of the trichogenic tumors.

We herein present a case of trichoblastoma which occurred on the scalp as an erythematous plaque in a 42-year-old man. Histologically, the tumor was composed of lobular aggregation of basaloid epithelial cells and a few keratin cysts. Immunohistochemical studies showed the diffuse positive immunolabelling with RCK108 (monoclonal antibody against CK19) and MNF116 (against CKs 5, 6, 8, 17 and probably 19). Based on these findings, we can infer that trichoblastoma differentiates mainly toward the outermost layer of the outer root sheath between the lower permanent portion and the upper transient portion as shown in previous studies. (Ann Dermatol 16(4) 180~184, 2004)

Key Words: Trichoblastoma, Immunohistochemistry, Cytokeratin

INTRODUCTION

Trichogenic tumors are uncommon cutaneous neoplasms of the hair germ cells. Headington[1] divided theses tumors into four categories (trichoblastoma, trichoblastic fibroma, trichogenic trichoblastoma and trichogenic myxoma) based on the degree of maturation and relative porportion of epithelial and mesenchymal components. In 1993, Ackerman et al.[2] proposed to use the term "trichoblastoma" as an inclusive term for all benign follicular germinative cell tumors because of the overlapping nature of trichogenic tumors. On the other hand, Czernobilsky et al.[3] classified a large, solitary and deeper tumor showing the similarity to the classical trichoepithelioma to "giant solitary trichoepithelioma". As trichogenic tumors demonstrated a variable range of histologic patterns, complete histopathologic definition and classification of these tumors are not established yet.

We herein report a case of trichoblastoma presented as an erythematous plaque on the scalp. Immunohistochemical studies, using specific cytokeratin (CK) markers, show the differentiation toward the outermost layer of the outer root sheath between the lower permanent portion and the upper transient portion.

CASE REPORT

A 42-year-old man presented with asymptomatic, solitary plaque on the scalp for 15 years. A physical examination revealed about 1 cm-diametered, erythematous plaque with rubbery consistency (Fig. 1). There was neither ulceration nor rolled border. The cervical lymph node was negative. His past and family history was not contributory.

Biopsy specimens showed the presence of a symmetrical tumor mass occupying dermal and upper

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subcutaneous tissue (Fig. 2A). The tumor was composed of nests and cords of basaloid cells with peripheral palisading nuclei, surrounded by fibrous stroma (Fig. 2B). A few keratin cysts were also seen. The retraction clefts were not found between tumor and surrounding stroma but between stromas. Immunohistochemical analysis was performed with monoclonal antibodies, MNF116 (against CKs 5, 6, 8, 17 and 19), 35βH11 (CK8), RCK108 (CK19), Ks20.8 (CK20), CD34 and vimentin. Diffuse positive immunolabelling was observed in the MNF116 preparation (Fig. 3A). The monoclonal antibody against CK19 decorated the epithelial cord and keratin cyst with peripheral accentuation (Fig. 3B). Monoclonal antibodies against CD34 and vimentin showed sparse and diffuse staining results in stromal cells, respectively. None of the tumor cells were immunolabelled with monoclonal antibody against CK8 and CK20.

On the basis of clinical and histological findings, we diagnosed this case as a trichoblastoma. We excised the tumor completely and no evidence of recurrence was observed for 6 months.

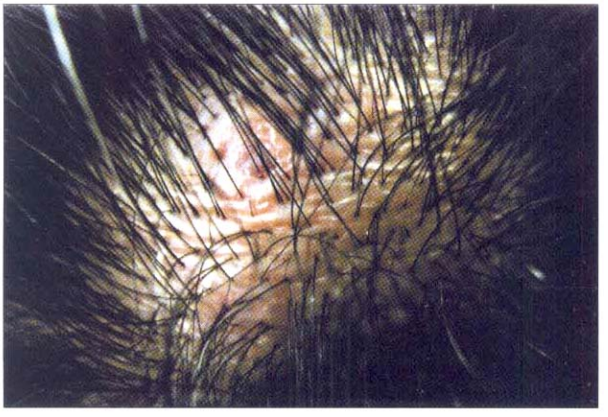
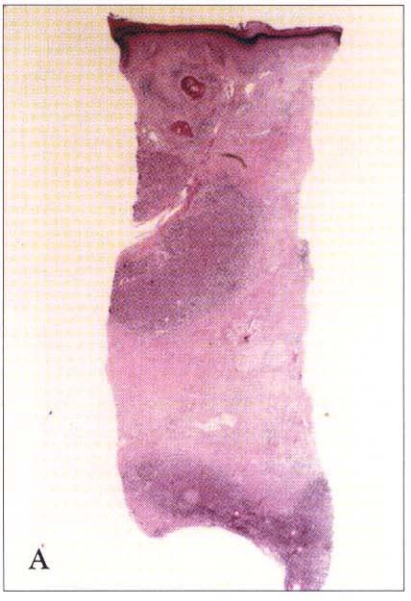


Fig. 1. Solitary, 1 cm-diametered, erythematous plaque with rubbery consistency on the scalp.

DISCUSSION

Trichogenic tumors are an uncommon cutaneous neoplasm originated from the hair germ cells. As these tumors demonstrated a variable range of histologic patterns, complete histopathologic definition and classification of them are very difficult and complex. In 1976, Headington[1] divided these tumors into four categories, based on the extent of stromal components and the degree of follicular differentiation; trichoblastoma, trichoblastic fibroma, trichogenic trichoblastoma and trichogenic myxoma. Trichoblastoma is primarily an epithelial tumor without stromal induction. Trichoblastic fibroma and trichogenic trichoblastoma show mixed epithelial-mesenchymal components and are subdivided by the degree of follicular differentiation. Mesenchymal trichogenic tumors are referred to as trichogenic myxoma. Because there are overlapping features in these subclasses and more than two subclasses are shown in the same tumors, Ackerman et al.[2] sug-



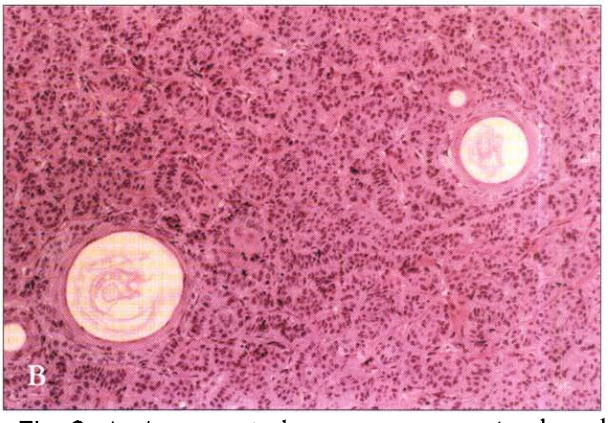


Fig. 2. A. Asymmetrical tumor mass occupies dermal and upper subcutaneous tissue. B. The tumor was composed of nests and cords of basaloid cells with a few keratin cysts (H&E, A. × 20, B. × 200).

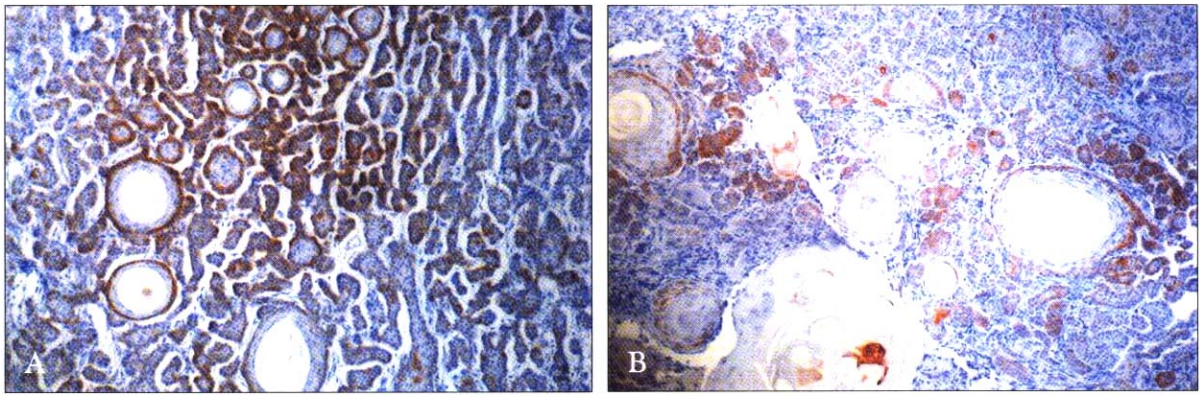


Fig. 3. A. Epitheloid cells are stained with MNF116 (monoclonal antibody against CKs 5, 6, 8, 17 and 19) B. tumor nests were immunolabelled with RCK108 (against CK19) with peripheral accentuation (A. MNF116, B. RCK108, × 100).

gested to use the comprehensive term "trichoblastoma" including all these groups of tumors and to subdivide them into large nodular, small nodular, cribriform, retiform and racemiform according to the cellular arrangement.

Trichoblastomas are usually presented as solitary, small, skin-colored nodules on the face and scalp without ulceration. They may occur primarily as dermal neoplasm or secondarily in association with sebaceous nevus[4]. Histologically, they are characterized by basaloid proliferation in which the neoplastic cells are arranged in cords, sheets, or discrete clusters surrounded by fibrous stroma. Hair differentiation is evident either as immature cellular clusters like primitive hair papilla and hair bulb or as more mature proliferative foci that resemble hair root structure[5]. According to Ackerman's classification, our case is classified as combined large nodular and small nodular form.

Expression of specific CK in the epithelial components varies between the stages of development and differentiation, therefore, the immunohistochemical study using a panel of monoclonal antibodies against specific CKs may provide evidence of cellular differentiation and morphologic and/or growth features in various epithelial tumors. The staining patterns of CKs in normal pilar structure and trichoblastomas including our case are summarized in Table 1 and 2, respectively. Our case showed positive staining results for CK19 and negative results for CK8 and CK20. CK19 has been detected in the outer root sheath (ORS) cells of the adult human hair follicle, with the greatest concentration observed in the bulge region[13]. In addition, Watanabe et al.[14] reported CK19 was noted in the outermost layer of the ORS between the lower permanent portion and the upper transient portion. Together with our staining patterns, the immunoreactivities of trichoblastoma indicate similarity to the outermost layer of the ORS, especially between the lower permanent portion and the upper transient portion of normal hair follicles, namely the bulge area. However, many immunohistochemical studies for trichoblastomas, including our data, have shown the various staining results[6,8-12]. As a dynamic structure, hair follicle changes according to the hair cycle, anagen, catagen, and telogen phases in which the architecture of the subinfundibular hair follicle is completely changed. In addition, CKs may be influenced by local factors. This could be the result of variables such as inflammation, certain cytokines, growth factors, or vitamins[9]. Finally, the different antibodies for the same CK can show different sensitivity and specificity. These facts can be thought to influence, in concert, the immunohistochemical staining results.

The differential diagnoses of trichoblastoma include keratotic basal cell carcinoma and trichoepithelioma. Basal cell carcinoma can be excluded by the absence of the artifact cleft separating tumor from stroma, mucinous stroma, mitosis and necrosis. Trichoepithelioma occurs as multiple or solitary lesions with a propensity for face and rarely exceeds 0.5 cm in size. Histologically, it consists of the cribriform aggregation of basaloid cells and multiple keratin cysts, usually confined to the superficial

Table 1. Immunohistochemical Stains in Normal Pilar Apparatus[6,7]

Antibody	Specificity	Infundibulum	ORS	IRS
34βB4 DE-K10	CK 1 CK 10	S+	-	-
AE3	CKs 1-8	+	-	-
KL1	CKs 1, 2, 5-8, 11, 14, 16-18	S+	I+ → -	-
34βE12	CKs 1, 5, 10, 14	+	+	-
215B8, 6B10 Ks13.1, KS1A3	CK 4 CK 13	-	-	+**
MNF116	CKs 5, 6, 8, 17, probably 19	O+	+	-
LP34	CK 5, 6, 18	+	+	-
RCK102	CK 5, 8	O+	+	-
OV-TL12/30, Ks7.18 35βH11 CAM5.2 Ks18.04 RCK108	CK 7 CK 8 CK 8, 18 CK 18 CK 19	-	-	-
4.1.18 170.2.14 Ks19.1	CK 8 CK 19 CK 19	-	O+ → -	-
AE1	CKs 10, 14-16, 19	O+	+	-
Involucrin	Involucrin	I+	I+	+**

ORS: outer root sheath beneath the infundibulum. IRS: inner root sheath. S+, I+, O+: positive staining was noted in suprabasal cells, inner layers and outer layers, respectively. →: staining intensity changes in the lower portion of the ORS. +**: positive staining in IRS before keratinization.
This table is modified form of the result of reference[6,7].

Table 2. Immunohistochemical Stains in Trichoblastomas

	MNF116	CK 8	CK19	CK20	CD34
Torii et al[6].(1)	+	+	O+	NI	NI
Schirren et al[8].(30)	NI	-	+(30)	-	+(21)
Kruzen et al[9].(13)	NI	+(13)	+(9)	NI	NI
Yamamoto et al[10].(1)	NI	NI	+	NI	NI
Misago et al[11].(1)	NI	+	+	NI	NI
Lee et al[12].(4)	+(4)	-	-	+(1)	-
Our case	+	-	+	-	±

( ): case number. O+: positive staining was noted in outer layers. NI: not investigated.

dermis. However, Ackerman et al.[2] took it as a type of trichoblastoma restricted to the dermis.

We present a case of trichoblastoma in a 42-year-old man. We can infer from immunohistochemistry that trichoblastoma may differentiate mainly toward ORS, especially to the outermost layer of the ORS between the lower permanent portion and the upper transient portion.

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