Rippled-Pattern Trichoblastoma of the Scalp: Case Report

Saçlı Derinin Dalgalı Paterndeki Trikoblastoması

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ABSTRACT Rippled-pattern trichoblastomas are rare, benign, trichogenic tumors characterized by alternating bands of basaloid epithelial cords and stroma. A 47-year-old male applied for a scalp mass that had grown in three years. On dermatologic examination an erythematous, moderately hard, ulcerated and bleeding tumoral nodule, measuring 3 x 2 cm was seen in the right frontoparietal region of the scalp. He had surgery under general anesthesia and the lesion was excised totally. Histopathologic examination of the excised surgical material disclosed a tumoral mass around a dilated hair follicle. The tumoral mass was composed of band-like nests of basaloid cells showing peripheral palisation in a fibrocellular stroma. An immunohistochemical examination was performed. A diagnosis of rippled pattern trichoblastoma was concluded on the ground of the histopathologic findings. Only a few cases of rippled-pattern trichoblastoma have been reported in the literature. Here we present a new case of rippled-pattern trichoblastoma.

Key Words: Scalp; immunohistochemistry; skin; skin neoplasms, hair follicle

ÖZET Dalgalı paterndeki trikoblastomalar, derinin bazaloid epitelial kord ve stromasının değişen bandları ile karakterize nadir görülen, benign ve trikojenik tümörleridir. Kırk yedi yaşında bir erkek hasta saçlı deride üç yılda büyümüş bir kitle ile başvurdu. Dermatolojik muayenesinde, kafa derisinin sağ frontoparietal bölgesinde eritematöz, orta sertlikte, 3 x 2 cm boyutlarında ölçülen, ülsere ve kanayan tümoral nodül mevcuttu. Hasta genel anestezi altında ameliyat edildi ve lezyon total olarak çıkarıldı. Cerrahi olarak çıkarılan materyalin histopatolojik incelemesi, genişlemiş bir saç follikülü etrafında gelişen tümoral kitleyi ortaya koydu. Tümoral kitle fibrosellüler stroma içinde çitle sınırlandırılmış tarzda periferal dağılım gösteren bazaloid hücre kümesi şeklinde bantlardan oluşuyordu. İmmünhistokimyasal inceleme yapıldı. Bu histopatolojik bulguların zemininde dalgalı paternde trikoblastoma tanısı kondu. Literatürde dalgalı paternde trikoblastoma olarak bildirilen çok az vaka vardır. Bu olgu sunumunda biz de ek bir dalgalı paternde trikoblastoma vakası sunduk.

Anahtar Kelimeler: Skalp; immünhistokimya; deri; deri neoplazmları, kıl folikülü

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Trichoblastomas are benign adnexal tumors arising from follicular germinative cells.[1] This tumor type is one of the two most common benign tumors arising in a nevus sebaceous.[2] Patients with nevus sebaceous develop mostly benign neoplasms with a frequency of 13.6% at 46.3 years on the average.[2] The two most common benign neoplasms developing in a nevus sebaceous are syringocystadenoma papilliferum and trichoblastoma.[2]

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Rippled-pattern trichoblastomas are rare tumors characterized by immature follicular epithelial germinative cells with alternating bands of basaloid epithelial cords and stroma that look like waves on low power microscopic examination and resemble Verocay bodies.[1,3] There are only eight cases of rippled-pattern trichoblastoma reported in the literature.[1,4-7,8]

CASE REPORT

A 47-year-old male patient applied with the complaint of a mass located in his scalp. He stated that the lesion had been present since birth but enlarged in the previous three years. The lesion necessitated oral antibiotic treatment due to self irritation and infection. On dermatologic examination, an erythematous, moderately hard, ulcerated and bleeding tumoral nodule, measuring 3 x 2 cm in size was seen in the right frontoparietal region of the scalp (Figure 1). He was operated on under general anesthesia and the lesion was excised totally including galea aponeurotica with 5 mm surgical margins. The resulting defect was reconstructed with a partial thickness skin graft. Postoperative period was uneventful and there was no tumor recurrence postoperatively at the sixth month control examination.

Histopathological examination of the excised surgical material disclosed a tumoral mass developed around a dilated hair follicle and infiltrated the surrounding tissue in a nodular budding fashion in a fibrocellular stroma under superficially ulcerated epidermis (Figure 2). The tumoral mass was composed of band-like nests of basaloid cells showing peripheral palisation which were reminiscent of Verocay bodies (Figure 3). Necrotic and cystic areas and mucine congestion were observed in the center of some of these nests (Figure 2, 3). The basaloid cells were elliptical in shape without any atypia and contained scant cytoplasm and basophilic nuclei. The nuclear chromatin was finely granular and identifiable nucleoli were rare. More than 10 mitotic figures/10 high power field and apoptotic bodies were observed. Sporadic papillary mesenchymal bodies were also detected in histopathological sections (Figure 4). No retraction

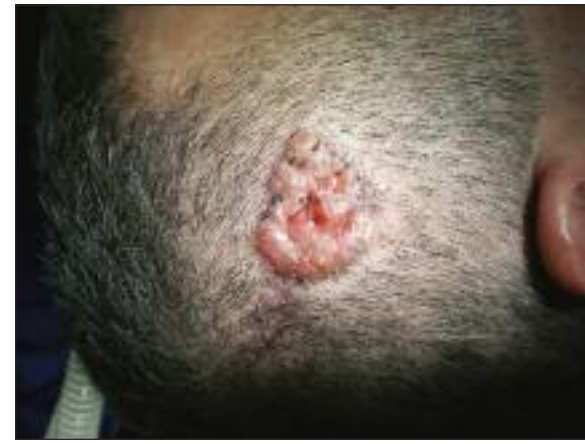

FIGURE 1: Clinical appearance of the patient: an erythematous, moderately hard tumoral nodule, measuring 3 x 2 cm is present in the right frontoparietal region.

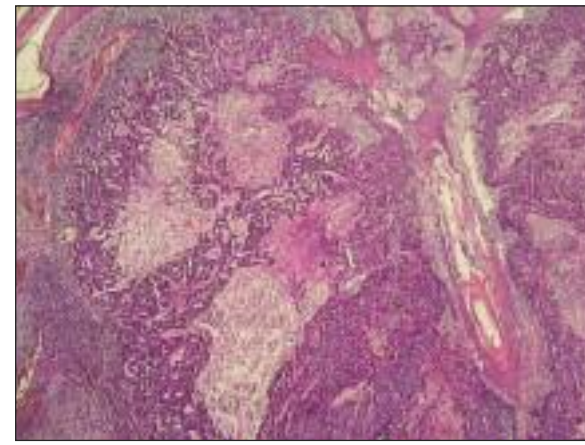

FIGURE 2: Tumoral mass is located in a fibrocellular stroma and centered around a dilated hair follicle, and it infiltrates the surrounding tissue in a nodular budding fashion. Necrotic and cystic areas are observed in the center of some of the tumoral islands (H&E, X40).

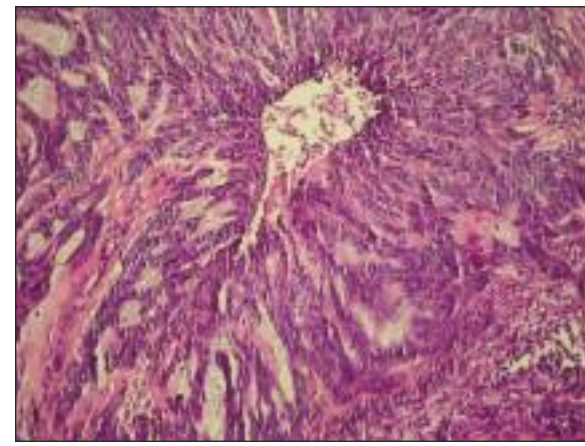

FIGURE 3: Band-like islands of basaloid cells showing central mucin congestion areas (H&E, X400).

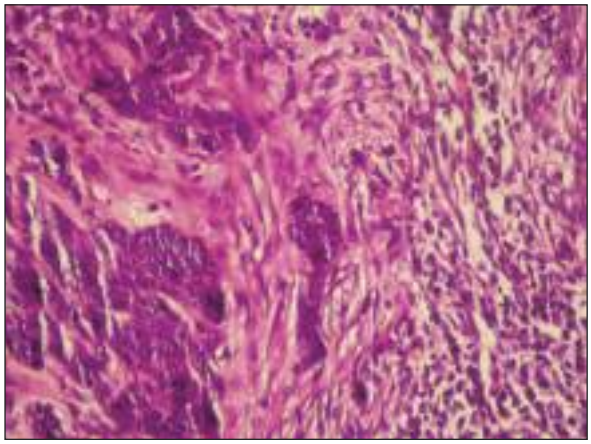

FIGURE 4: A papillary mesenchymal body (H&E, X400).

clefts were identified between tumor lobules and surrounding tumor stroma. An immunohistochemical examination was also performed. HMWK and panCK were found to be diffusely positive. CK7, CK8-18, and Ki-67 were stained as positive focally (Figure 5). However bcl2, CK20, CD34, EMA and p53 were found to be negative. A diagnosis of rippled-pattern trichoblastoma was concluded on the ground of these histopathological findings.

DISCUSSION

Trichoblastomas are benign adnexal tumors which consist of an epithelial component of follicular germinative cells and a stromal component. Tumor cells may form primitive hair papillae. Trichoblastomas may show five histologic patterns as small nodular, large nodular, cribriform, racemiform and retiform. However, one or more histological pattern can be seen in the same tumor.[6]

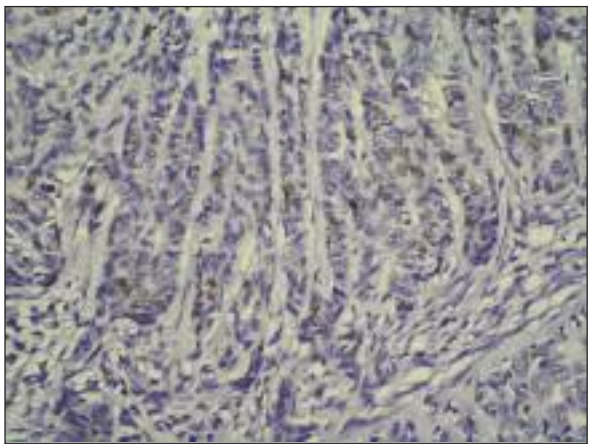

FIGURE 5: Ki-67, which is a proliferation marker indicating mitotic activity, was detected to be >10% positive (immunohistochemistry, X400).

Hashimoto et al.[7] were the first to describe a rare trichogenic tumor which they named "rippled-pattern trichomatricoma" in 1989. They have preferred the term "matricoma" to define this immature hair tumor or trichogenic tumor because the term "blastoma" implies a malignant potential.[7] However, the tumor that they reported as "rippled trichogenic tumor" is now considered as a variant of trichoblastoma.[4] Several characteristic features of this tumor have been described as proliferation of immature pilar cortical cells and a peculiar arrangement of tumor cells that resembles a plowed field.[7] In this tumor alternating bands of epithelial cords and stroma produce a rippled-pattern.[7]

Requena and Barat[6] reported in 1993 a giant trichoblastoma of the scalp showing different histopathological patterns as rippled, adenoid and palisading. These patterns were located in different tumor regions and the tumor showed sebaceous differentiation in some areas.[6] They have concluded that trichoblastoma is mainly composed of follicular germinative cells.[6] Six other cases of rippled-pattern trichoblastoma were reported in the literature.[1,4,5,8] The clinical and demographic features of eight cases reported in the literature and those of our case are presented in Table 1. The histopathological and immunohistochemical findings are shown in Table 2.

Kaddu et al.[9] reported that average age of the patients with solitary trichoblastoma was 51.7 years and male/female ratio was 11/24 (0.46). These lesions were solitary papules or nodules, measuring 0.5-8 cm in size and located in the head and neck region in 74.29% of cases.[9] The average age of reported rippled pattern trichoblastoma patients was 50.56 years and male/female ratio was 2 (Table 1). The tumors arose in Caucasians in 66.67% of the cases and the tumor was present for 7.41 years on the average (Table 1). Reported rippled-pattern trichoblastomas were usually asymptomatic and located mostly in the scalp in 88.89% of the cases (Table 1). Reported tumor nodule diameters ran-

TABLE 1: Clinical and demographic findings of rippled-pattern trichoblastomas.[1,4-8]

Author	Age	Sex	Race	Duration	Location	Symptom	Size	Lesion
Hashimoto (1989)	30	F	Black	9 years	Right cheek	-	2.5-3 cm	Ulcerated nodule
Requena & Barat (1993)	69	M	Caucasian	≥ 20 years	Scalp (vertex)	-	8 cm	Spherical mass
Akasaka (1997)	36	M	Asian	2 years	Scalp (left temporal)	-	12 mm	Alopecic ulcerated nodule
Yamamoto(2000)	62	F	Asian	5 years	Scalp (occipital)	-	10x8 mm	Irregular shaped nodule
Graham(2000)	67	M	Caucasian	3 months	Upper neck-Posterior hairline	-	8 mm	Pearly plaque
	58	F	Caucasian	10 years	Scalp vertex)	Easy bleeding & traumatization	1.5x1.0 cm	Verrucous plaque
	50	M	Caucasian	10 years	Scalp (occipital)	Slightly tender	6x8 mm	Verrucous papillomatous nodule
Swick (2009)	36	M	Caucasian	36 years (life long) with recent growth	Retroauricular scalp at superior pole of hairless brownish-yellow linear plaque	-	0.6 cm	Mamillated papular projection
Present case	47	M	Caucasian	47 years (life long) with growth in the last 3 years	Scalp (right frontoparietal)	Infection Bleeding	3x2 cm	Ulcerated and bleeding nodule

TABLE 2: The histopathological and immunohistochemical findings of rippled-pattern trichoblastomas.[1,4-8]

	Histopathology	Immunohistochemistry		
Author	Concomitant findings	Positive	Slightly/Focal positive	Negative
Hashimoto (1989)	Desmoplastic trichoepithelioma	HKN6	EKH4, AE1&3,S-100, MEL5, CD1	EKH5, EKH6, PAS, CEA, NSE,
Requena & Barat (1993)	Sebaceous differentiation	-	CAM5.2, Vimentin, S-100	CEA, EMA
Akasaka (1997)	Peripherally attached dilated follicular sheath Necrosis	Keratin	-	CEA, NSE, PAS, S-100
Yamamoto(2000)	Surrounding nevus sebaceous	CK1/5/10/14, CK14, CK7	CK10, CK10/11, CK5/8, CK17, CK19, S-100, Vimentin	CK2e, CK6, CK13, CEA, EMA, SMA
Graham (2000)	Sebaceous differentiation & sebaceous ducts	EMA	-	-
	Sebaceous differentiation	EMA	-	-
	Sebaceous differentiation	EMA	-	-
Swick (2009)	Apocrine differentiation	-	CK20, bcl-2, CD34, EMA	-
Present case	Dilated hair follicle Necrotic and cystic areas Mucine congestion areas	panCK, HMWK	CK7, CK8-18, Ki-67	CK20, CD34, EMA, bcl2, p53

ged from 6 mm to 8 cm, and the average diameter of the tumor nodules was 21.33 mm (Table 1).

Swick et al.[8] recently reported a case of rippled-pattern trichoblastoma arising in a nevus sebaceous. They reported a superficial and deeper dermal involvement and superficial location of trichoblastomas is a characteristic of development in nevus sebaceous.[8] The tumor in our case could have developed in a previous nevus sevaceous since the history of the lesion goes back to birth. However we failed to find any histopathological findings suggestive of nevus sebaceous. However, this does not exclude the possibility that the lesion developed in a nevus sebaceous because the tumoral lesion was superficially located with an epidermal connection.

Our histopathological findings are similar to those reported in other studies. The tumoral mass was composed of band-like nests of basaloid cells which were reminiscents of Verocay bodies seen in neurilemmoma. We have also observed that the tumor nests seem to sprout out from a dilated hair follicle and they contain necrotic, cystic and mucine congestion areas. Immunohistochemically, we did not detect CK20 positivity in our rippled-pattern trichoblastoma (Table 2). CK20 has been re-

ported to be positive in 70% of trichoblastomas and this marker indicates Merkel cell differentiation.[10] CK20 staining is scarce to absent in basal celle carcinoma (BCC).[8] Stromal staining of CD34 has been found to be positive in 40% of trichoblastomas in the stroma adjacent to the tumor lobules but staining with this marker was negative in our case.[8,10] BCCs fail to demonstrate staining with this marker.[8] These findings may suggest that rippled-pattern trichoblastomas may be immunohistochemically somewhat different from other trichoblastomas (Table 2). Bcl-2 is a proliferation marker and has been found to be positive in 100% of BCCs and has been found only at the periphery of the tumor nests in trichoblastomas.[8,10] However, staining with this marker was negative in our case. Thus negative staining with this marker helps differentiating benign trichoblastoma from BCC.[10] Swick et al.[8] recently reported a case of rippled-pattern trichoblastoma showing a different immunohistochemistry profile with focal positivity of CK20, bcl-2 and CD34 in comparison with our case. Staining with p53 was negative in our case. This is an apoptosis marker and negative staining with this marker indicates benign nature of our tumor. However, p53 has been detected in trichoblastic sarcoma.[11] Ki-67 is a proliferation marker indicating mitotic activity and it was detected more than >10% positive in our case. It has been found negative in basal cell carcinomas.[11]

Trichoblastomas are benign hair germ tumors. Rippled-pattern trichoblastomas are rare tumors characterized by alternating bands of basaloid epithelial cords and stroma resembling neurilemmomas. In this case report, we report an additional case of rippled-pattern trichoblastoma for which immunohistochemical studies were performed. We also reviewed other very few rippled-pattern trichoblastoma cases reported in the literature.

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