Acta Derm Venereol 2014; 94: 114–115

SHORT COMMUNICATION

Tubular Spitz Naevus Mimicking Eccrine Spiradenoma

Yasuyuki Fujita[1], Satoru Aoyagi[1], Masumi Tsujiwaki[1], Erina Homma[1], Hideyuki Ujiie[1], Hiromi Fujita[2], Kanako C. Hatanaka[2] and Hiroshi Shimizu[1]

[1]Department of Dermatology, Hokkaido University Graduate School of Medicine, North 15 West 7, Kita-ku, Sapporo 060-8638 and [2]Department of Surgical pathology, Hokkaido University Hospital, Sapporo, Japan. E-mail: yfujita@med.hokudai.ac.jp

Accepted Mar 25, 2013, Epub ahead of print Jun 5, 2013

Spitz naevus was first described by Sophie Spitz in 1948 (1). It is a variant of benign melanocytic naevus composed of spindled and epithelioid melanocytes, found predominantly in children and adolescents. A number of histopathological variants have been reported, such as desmoplastic, hyalinized, angiomatous, granulomatous, pagetoid, myxoid, tubular and rosette forms (2). We report here a case of tubular Spitz naevus with variable histopathological images, which partly resembled ependymal rosette in neurogenic neoplasms and partly resembled eccrine spiradenoma.

CASE REPORT

A 9-year-old boy presented with an asymptomatic reddish nodule, which had been present for 2 months. On clinical examination, a pink-red, elastic hard nodule with papillomatous surface, 8 mm in diameter, was noted on his left forearm (Fig. 1). He was otherwise healthy, and no other specific skin eruptions were found. Pseudolymphoma, xanthogranuloma and Spitz naevus were considered clinically. Since the eruption had not changed over a 3-month period careful observation, we excised it for histopathological diagnosis. In the dermoepidermal junction, spindle cells with faint melanins were aggregated to form nests (Fig. 2a). Small nests of tumour cells showed some clefts (Fig. 2b). Giant cells and dense lymphocytic infiltration were also observed. In the entire dermis, round-to-oval epithelioid cells with eosinophilic cytoplasm were remarkably proliferated. Interestingly, the majority of such tumour cells formed microtubular appearance, variably mixed with microvenules (Fig. 2c). A mass of basophilic tumour cells with microcystic appearance was also observed in the subcutis, mimicking eccrine spiradenoma (Fig. 2d). Regardless of the diverse appearance of tumour cells, these cells had a uniform immunohistochemical profile; S-100^{+}, Melan-A^{+}, HMB-45^{-} and AE3/AE1^{-} (Fig. 2e and f). From these clinicopathological findings we diagnosed the patient with tubular Spitz naevus. After excision no local recurrence or metastasis has been observed to date.

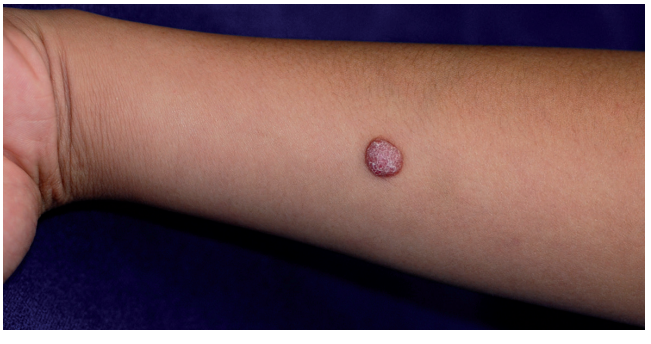

Fig. 1. A pink-red, elastic, hard, asymptomatic nodule with papillomatous surface on the left forearm.

DISCUSSION

Tubular Spitz naevus, a very rare variant, was first described by Burg and colleagues in 1998 (3). They reported a case with a characteristic finding, the formation of tubular structures bordered by S-100^{+} epithelioid cell strands. Confocal laser scan microscopic analysis revealed tubular and/or microcystic spaces formed by tumour cells, at least 30 μm long. Requena et al. (2) investigated 349 lesions of Spitz naevi, and detected only one tubular type case.

The pathomechanisms and clinical significance of tubular Spitz naevus are unknown. Burg et al. (3) speculated that the tubular images might be caused by apoptosis of centrally located naevus cells or secretion of autocrine or paracrine factors. Conversely, Ziemer et al. (4) reviewed 31 Spitz naevi and found "tubular" structures in 15 cases, and they suggested that such tubular appearance may result from retraction of aggregated epithelioid tumour cells, secondary to fixation of formalin. As a similar histopathological variant, Kantrow and colleagues (5) reported a case of Spitz naevus with rosette-like formation. They pointed out that the rosettes resembled Homer-Wright rosettes in neurogenic tumours, such as neuroblastoma, characterized by a radial arrangement of cells with centrally situated fibrillar material composed of neuropil. In our case, unlike Homer-Wright rosettes, tumour structures contained apparent empty-appearing lumen and lacked fibre-rich neuropil or central cytoplasmic projections. Such tubular patterns partly resembled "true ependymal rosette" observed in well-differentiated ependymoma (6). Kantrow et al. (5) speculated that shared neural crest origin might induce similar characteristic images to neurogenic tumours, which also seemed to explain our case. Indeed, rosette-like, tubular or glandular patterns are observed in other melanocytic neoplasms, such as

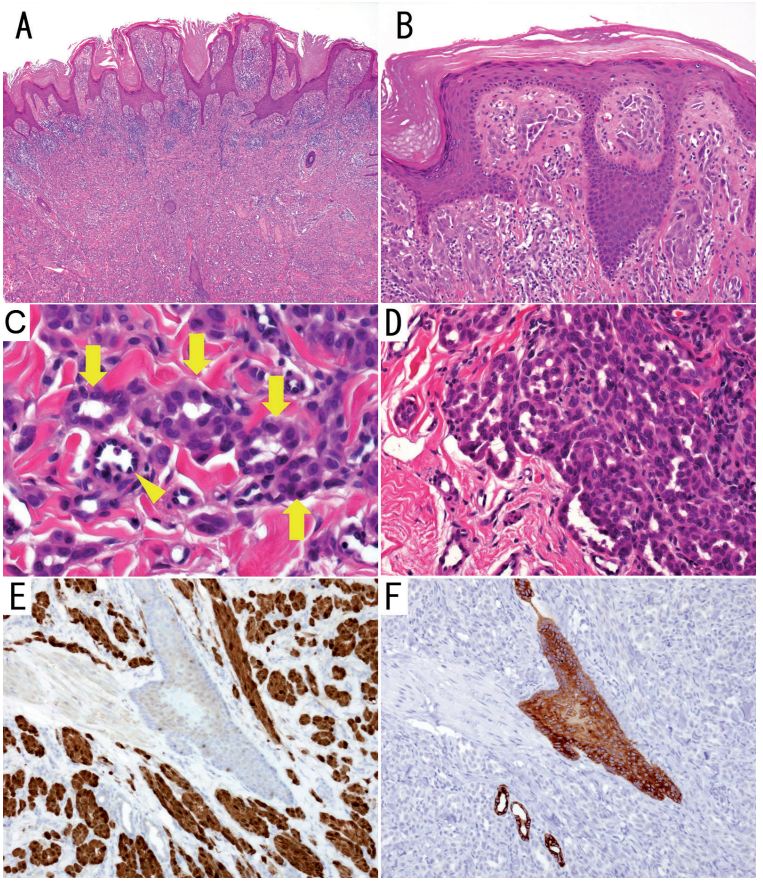

Fig. 2. Histopathological findings. (a) Tumour cells were proliferated in the entire dermis, and nests of tumour cells with hyperkeratosis were formed in the dermo-epidermal junction (haematoxylin and eosin (H&E) ×40). (b) Small nests of tumour cells demonstrated clefts, probably formed by artefacts from formalin fixation (H&E ×150). (c) Round-to-oval epithelioid cells with prominent tubular/rosette-like appearance were observed (*arrows*). Note microvenules with neutrophil in the lumen (*arrowhead*) (H&E ×400). (d) Dense basophilic cells mimicking eccrine spiradenoma were also observed in the subcutis (H&E ×200). (e) The tubular tumour cells showed positive S-100 (×200). (f) AE1/AE3 was negative. Note positivity in the normal sweat ducts (×200).

Clark's naevus and malignant melanoma (7, 8). Taking these observations into consideration, tubular, rosette and glandular structures seen in Spitz naevus might reflect declined differentiation to neurological lineages from neural crest, and the degree of artefacts caused by formalin fixation can explain histopathological differences in these structures.

Since dense basophilic tubular structures were observed in the subcutis mimicking eccrine spiradenoma, we first considered the case might be Spitz naevus with sweat duct differentiation or concomitant sweat duct tumours, such as spiradenoma and eccrine angiomatous hamartoma. Recently, a case of Spitz naevus in concordance with a syringoma was reported, which might support a possible co-occurrence of Spitz naevus and eccrine duct neoplasms (9). However, in our case, the positivity of S-100 and negativity of AE1/AE3 in the tubular areas denied the concomitant sweat gland neoplasms. Although the significance of the rare histological subtype is unknown, dermatologists and pathologists should avoid potential misdiagnosis caused by the wide variety of histopathological images in Spitz naevus.

The authors declare no conflicts of interest.

REFERENCES

1. Spitz S. Melanomas of childhood. Am J Pathol 1948; 24: 591–609.
2. Requena C, Requena L, Kutzner H, Sánchez Yus E. Spitz nevus: a clinicopathological study of 349 cases. Am J Dermatopathol 2009; 31: 107–116.
3. Burg G, Kempf W, Höchli M, Huwyler T, Panizzon RG. ''Tubular'' epithelioid cell nevus: a new variant of Spitz's nevus. J Cutan Pathol 1998; 25: 475–478.
4. Ziemer M, Diaz-Cascajo C, Kohler G, Weyers W. ''Tubular Spitz's nevus'' an artifact of fixation? J Cutan Pathol 2000; 27: 500–504.
5. Kantrow S, Kalemeris GC, Pietro V. Spitz nevus with rosette-like structures: a new histologic variant. J Cutan Pathol 2008: 35: 510–512.
6. Wippold FJ 2nd, Perry A. Neuropathology for the neuroradiologist: rosettes and pseudorosettes. AJNR Am J Neuroradiol 2006; 27: 488–492.
7. Soyer HP, Breier F, Cerroni L, Kerl H. 'Tubular' structures within melanocytic proliferations: a distinctive morphologic finding not restricted to Spitz nevi. J Cutan Pathol 1999; 26: 315–317.
8. Tarlow MM, Nemlick AS, Rothenberg J, Schwartz RA. Pseudoglandular-type melanoma: a rare melanoma variant. J Cutan Pathol 2008; 35: 588–590.
9. Piana S, Ragazzi M, Zalaudek I, Argenziano G. A curious serendipitous finding: Spitz naevus combined with a syringoma. Australas J Dermatol 2013; 54: e64–66.