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A Case of Eccrine Spiradenoma: A Rarely Seen Soft Tissue Tumor on the Extensor Surface of Arm

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Dear Editor:

A 40-year-old man presented with a nodule on the extensor surface of his left forearm. He couldn't remember when it formed. Initially, it showed no symptoms but with time, became painful. Physical examination revealed a slightly bluish to skin colored, hard mobile nodule on the left forearm measuring 1×1 cm in size (Fig. 1A). The initial clinical suspicion was epidermal cyst, trichilemmal cyst, or lipoma; thus, ultrasonography (US) was done. US results indicated a well-defined oval isoechoic mass in the subcutaneous fat layer, with moderate hyperemia (Fig. 1B). A complicated epidermoid cyst, or other hypervascular mass, such as vascularized leiomyoma or (least likely) a sarcoma (e.g., malignant fibrous histiocytoma) was suspected. Excisional biopsy was performed. Microscopic examination showed a deep, well-defined tumor with basophilic lobules, encapsulated by thick connective tissue in the dermis and subcutaneous layer. A trabecular arrangement of basophilic cells was present (Fig. 1C). Two types of cells (small, dark, basaloid cells with hyperchromatic nuclei, and cells with large, pale, ovoid nuclei) were seen (Fig. 1D). The diagnosis was confirmed as eccrine spiradenoma (ES). The tumor was completely excised.

ES is a rare benign soft tissue tumor first described by Kersting and Helwig (1956). It may occur at any age, but typically affects young adults aged 15～35 years, with no sexual predilection[1]. It presents as a solitary mass localized in the skin and subcutaneous tissue, smaller than 1 cm in size. Paroxysmal pain and tenderness may often present[2]. It tends to arise on the upper part of the body; head, neck, and trunk, especially ventral portion[1]. Rarely, it may be located on the upper and lower extremities, especially dorsal portion[3]. The summarization of the previously reported cases on extremities was described in Table 1. ES lesions are mostly solitary, but rarely multiple, linear, blaschkoid, or grouped. The pathogenesis is thought to be related to differentiation, mainly of the secretory portion of the secretory coil of eccrine sweat glands[2,4]. The presence of myoepithelial cells and phosphorylase, demonstrate the eccrine origin of the tumor. However, several evidences suggest its origin is apocrine rather than eccrine[4]. It is sometimes associated with trichoepithelioma and cylindroma, tumors of apocrine or folliculosebaceous-apocrine origin. US study revealed a well-demarcated mass with lobulated contours, which didn't have any relation with the epidermis and didn't extend into the neighboring muscular tissues. It was localized in the superficial part of the subcutaneous tissue[3,5]. When the mass shows well-defined hypoechogenicity, hypervascularity, no cystic portion, no hypoechoic scar, and when it is deeply located, an ES can be highly considered[5]. The essential histologic features include sharply demarcated nodules of basaloid cells in the dermis or in subcutaneous tissue (blue balls)[5]. Basaloid cells are composed of two types which might not be so apparent: one is paler,

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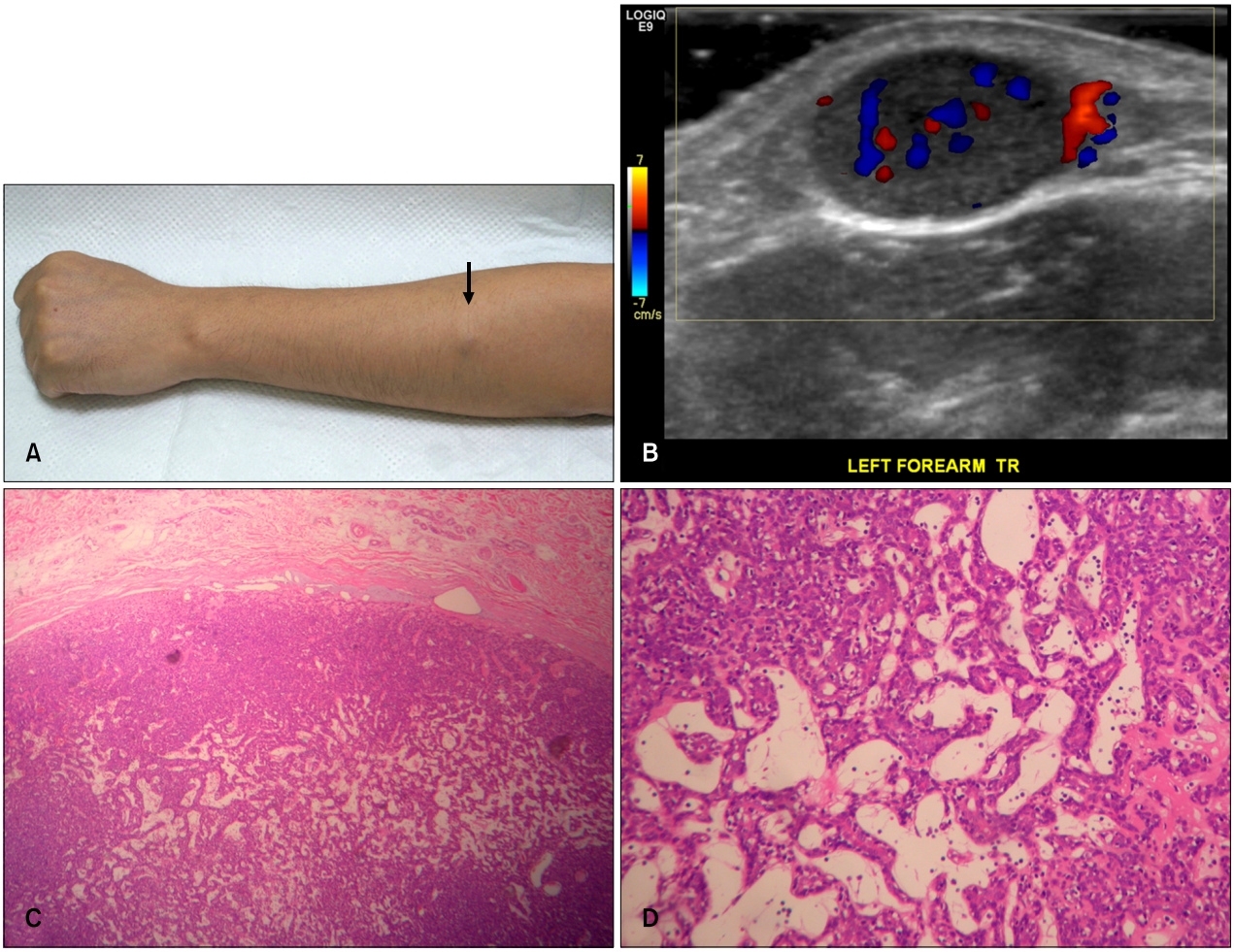

Fig. 1. (A) A solitary, mild bluish to skin-colored, hard mobile nodule on the extensor surface of the left forearm measuring 1×1 cm (black arrow). (B) Using ultrasonography, a well-defined oval isoechoic mass of about 10×7×10 mm, with moderate hyperemia, was determined in the subcutaneous fat layer on the left forearm. (C) Deep well-defined multinodular tumor with basophilic lobules encapsulated by thick connective tissue in the dermis and subcutaneous layer. A trabecular arrangement or clusters of deep basophilic cells were present (H&E, ×40). (D) In the nodule, two types of cells (small, dark, basaloid cells with hyperchromatic nuclei, and cells with large, pale, vesicular, and ovoid nuclei) were seen. The center of the lesions was composed of pale cells and duct like structures. The mitotic activity was low and there was no necrosis (H&E, ×200).

Table 1. Reported cases with eccrine spiradenoma developed on extremities

Reports	Age (yr)/ sex	Location	Characteristic finding	Pathologic diagnosis; imaging findings	Treatment
Alfonso-Trujillo et al. (2009)	17/F	Leg	0.3 to 0.5 cm sized, tender, nodular lesions of 7 years' duration. Zosteriform pattern	Eccrine spiradenoma; -	Surgical excision
Altinyazar et al. (2003)	32/F	Leg	Multiple, skin-colored, grouped, firm, tender, papulonodular lesions of 2 years' duration. Zostiform pattern	Eccrine spiradenoma; -	-
Bedlow et al. (1999)	19/F	Multiple (Rt. arm, Rt. leg, Rt. side of trunk)	15 year's duration, Tender blue-red dermal and subcutaneous nodules. Linear	Eccrine spiradenoma; -	-
Blanchard et al. (1981)	24/F	Leg	0.2 to 1 cm sized nodules for almost 12 years. Linear pattern. Asymptomatic	Eccrine spiradenoma (Linear eccrine nevus with comedones); -	-
Bourrat et al. (1992)	16/F	Arm, shoulder	Segmental lesions with the pattern of Blaschko's lines since 5 years ago.	Eccrine spiradenoma; -	-

Table 1. Continued

Reports	Age (yr)/ sex	Location	Characteristic finding	Pathologic diagnosis; imaging findings	Treatment
Brahim et al. (2009)	75/M	Rt. shoulder	A well-defined, motile, firm 5.0×4.0 cm sized nodule. Tender in pressure. A 6-year history of a recurrent mass that was removed 2 years ago	Eccrine spiradenoma, recurrent malignant eccrine spiradenoma; -	Surgical resection
Braun-Falco et al. (2003)	23/F	Multiple (Lt. lower leg)	1.5 cm sized multiple nodules almost covering the frontal aspects of lower leg for almost 8 years without obvious change. Linear. Occasionally tender	Eccrine spiradenoma (focal malignant transformation); -	Surgical resection
Criton et al. (1996)	11/M	Multiple (Lt. arm, Lt. leg, chest)	0.4 to 1.0 cm sized painful tender skin colored papule of duration of 4 years. Zostiform pattern	Eccrine spiradenoma; -	-
Englander et al. (2011)	55/M	Multiple (Rt. chest, Rt. arm)	0.5 to 6.0 cm sized multiple, well-circumscribed, subcutaneous, blue-grey nodules in dermatomal distribution of 20 years' duration. Painful	Eccrine spiradenoma; -	Surgical resection
Fahy et al. (1987)	37/F	Rt. hand	A 3.5×2.0 cm sized firm, non-tender swelling, of two years' duration, situated just distal to the right anatomical snuff-box	Eccrine spiradenoma; -	Surgical excision
Han et al. (2007)	24/F	Multiple (face, neck, chest, extremities)	0.3～2.0 cm sized well-defined nodules. Zosteriform. Painless	Eccrine spiradenoma; MRI, low SI in T1WI, high SI in STIR	-
Hashimoto et al. (1966)	15/F	Forearm	Segmental patterned lesions with 7 years' duration. Asymptomatic	Eccrine spiradenoma; -	-
Hemalatha et al. (2015)	80/F	Rt. upper thigh	A 1.75×0.75 cm sized painless raised blue to blackish lesion on thigh. Non-tender with smooth surface	Eccrine spiradenoma; -	Surgical excision
Ikeya et al. (1987)	37/F	Face, trunk, arm	0.1 to 0.8 cm sized multiple papular lesions of 30 years' duration. Linear pattern	Eccrine spiradenoma; -	-
Jin et al. (2008)	34/F	Lt. upper arm	A 1.8×1.7×1.1 cm sized firm nodular mass on the distal portion of the left upper arm, of approximately 5 years' duration. Painful	Eccrine spiradenoma; US, well-defined lobulating mass, vascularity(+), heterogeneous hypoechogenicity	Surgical excision
Laura et al. (2011)	55/M	Multiple (Rt. arm, Rt. chest)	0.5 to 6 cm sized multiple, well-circumscribed, subcutaneous, blue-grey nodules of 20 years' duration. Painful	Eccrine spiradenoma; -	Surgical excision
Martinez et al. (1992)	48/F	Multiple (Lt. leg)	0.2 to 0.6 cm sized multiple papulonodular blue-pink colored lesions appeared 18 years earlier. Asymptomatic. Tender with a firm, elastic consistency	Multiple linear cylindromas overlapping features with eccrine spiradenoma; -	Regular follow-up
Nath et al. (2009)	14/M	Rt. lower leg	A 0.5 to 1.0 cm sized multiple nodules on the posterior aspect of the entire length of the right lower limb since 2 years of age. Linear pattern (Blashkoid distribution). Painful	Eccrine spiradenoma with chondroid syringoma; US, dermal tumors, normal vessels of the lower limb, no connection with the underlying vessels and the tumors	Surgical excision

Table 1. Continued

Reports	Age (yr)/ sex	Location	Characteristic finding	Pathologic diagnosis; imaging findings	Treatment
Noto et al. (1994)	16/F	Multiple (Rt. leg, Rt. face, Rt. neck, Rt. trunk)	0.5 to 5.0 cm sized nodules, observed since birth. Bluegray to bright red colored painful papulonodular lesions. Linear nevoid pattern. Painful	Linear eccrine spiradenoma; -	-
Ohtsuka et al. (2002)	47F	Multiple (Rt. finger, Rt. hand, Rt. forearm)	0.7 to 2.0 cm sized linear, nontender lesions of 30 years' duration. Localized pattern	Eccrine spiradenoma; -	Surgical excision
Shaikh-Naidu et al. (2003)	34/F	Multiple (Rt. forearm)	10×2 cm area of 12 subcutaneous nodules with overlying cutaneous scar and bluish discoloration. Previously sustained a superficial abrasion. Linear. Mild discomfort	Eccrine spiradenoma; -	Surgical excision
Siegel et al. (2008)	40/M	Rt. hand	A 6.0×4.0 cm sized slightly mobile, painless mass of 20 years' duration. No lymphadenopathy	Giant eccrine spiradenoma; MRI, high heterogenous SI in T1WI	Surgical resection
Tsur et al. (1981)	35/M	Multiple (Lt. arm)	1 to 5 cm size. 26 years of duration. Bluish and shiny with congested blood vessels. Some tumors attached by short broad stalk. Linear pattern. Painful	Eccrine spiradenoma; -	Surgical resection
Yamakoshi et al. (2008)	76/M	Rt. upper arm	A 5.0×3.4×2.6 cm sized pale red pedunculated tumor with erosive surface. No symptom except for bleeding	Giant vascular eccrine spiradenoma; Enhanced CT, high-density nodules in the peripheral region and the central region with low density and no enhancement	Surgical excision

F: female, M: male, Rt.: right, Lt.: left, -: absent or not available, MRI: magnetic resonance imagning, SI: signal intensity, WI: weighted image, STIR: short tau inversion recovery, US: ultrasonography, CT: computed tomography.

with more cytoplasm than in the darker cells[1]. Malignant change is rare[2]. However, in cases with incomplete surgical removal, a high risk of local recurrence has been reported[2,5]. Early and complete surgical excision is the diagnostic tool and curative treatment option for ES[2].

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CONFLICTS OF INTEREST

The authors have nothing to disclose.

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