Linear Syringocystadenoma Papilliferum of the Arm: A Rare Localization of an Uncommon Tumour

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Sir,
Syringocystadenoma papilliferum (SCAP) is a rare benign adnexal skin tumour of apocrine or apoeccrine type with characteristic histopathological features, and varied and non-distinct clinical findings (1). The vast majority of lesions are solitary and occur in the head and neck region. Their occurrence at other anatomical sites is uncommon, and linear arrangements of these lesions are particularly rare (2). We report here an unusual case of multiple SCAP in a linear distribution presenting on the arm.

CASE REPORT

An otherwise healthy 19-year-old woman presented with papules present since birth. Physical examination revealed multiple linear arranged, discrete, erythematous, 0,5–1 cm sized pseudovesicular papules on the extensor site of proximal part of right upper extremity, close to the shoulder (Fig. 1a). Routine laboratory investigations were within normal limits. Histopathological examination of punch biopsy specimens taken from the lesions showed epidermal invaginations lined by a stratified epithelium in the superficial portions and double layered rows with basal cuboidal cells and luminal columnar cells in the lower portions. Papillary projections protruding into the lumen and within a fibrovascular stroma containing large numbers of plasma cells were present. There were no epidermal hyperplasia, nor abnormal hair follicles, or sebaceous glands (Fig. 1b). The diagnosis was consistent with SCAP. The lesion was totally excised, and the histopathology revealed SCAP.

DISCUSSION

SCAP is a sweat gland tumour that is not clinically distinct; a biopsy is usually required for diagnosis. Two different primary lesions have been described: a solitary plaque or one to several papules. The plaques are usually less than 4 cm in diameter and skin-coloured to dark brown. They may be flat and smooth, or raised with a papillomatous or verrucous surface. The less common papular lesions are skin-coloured to pink and less than 1 cm in diameter (1).

SCAP is usually observed as a warty plaque most commonly located on the head or neck region, where it may occur *de novo* or within a naevus sebaceous (1–3). However, it was also reported that 20% of lesions occurred on the trunk and 5% on the extremities, almost all on the lower extremities (3). Other unusual locations have included the breast (4, 5), buttock (6), inguinal and perianal regions, and scrotum (2) and on a postoperative scar (7). In about half of cases the lesions are present at birth (1, 3). As far as we know, there has been only one case report published previously with upper extremity involvement (8). Our case is the second reported case of SCAP on the arm.

As far as we have observed, there have been only 8 previous cases of linear SCAP reported in the literature in English (Table I) (2, 3, 8–13). The most recent of

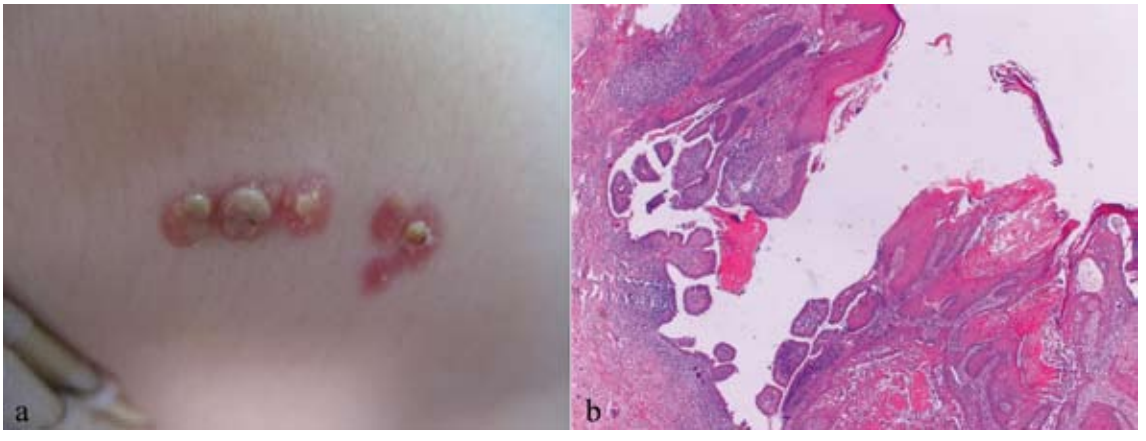

Fig. 1. (a) Multiple linear, discrete, erythematous, pseudovesicular papules on the extensor site of proximal part of right upper extremity. (b) Epidermal cystic invagination of epidermis to dermis, and papillary projections towards the lumen (haematoxylin and eosin; H&E ×40).

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Table I. *Cases of linear-arranged syringocystadenoma papilliferum reported in the literature*

Reference	Age at diagnosis (years)/age at onset	Gender	Location
Rostan & Waller (8), 1976	10/birth	Female	Upper extremity
Goldberg & Esterly (11), 1985	6/birth	Male	Neck
Premalatha et al. (10), 1985	16/birth	Female	Trunk
Epstein et al. (12), 1990	12/birth	Male	Trunk
de Bliek & Starink (3), 1999	11/birth	Female	Lower extremity
Patterson et al. (2), 2001	14/early childhood	Female	Lower extremity
Dawn & Gupta (13), 2002	?*	Female	Neck
Laxmisha et al. (9), 2007	5/6 months	Female	Scalp, neck
Present case	30/birth	Female	Upper extremity

*Data not available.

these occurred on the scalp and neck (9, 13) and the lower extremity (2).

It appears that SCAP can present rarely as multiple lesions in a linear array, unassociated with a naevus sebaceous or an epidermal naevus. We therefore agree with the suggestion of Patterson et al. (2) that SCAP should be included among the other adnexal tumours that are capable of forming linear arrangements on their own.

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