Letters to the Editor

Sir,

Syringocystadenoma papilliferum (SCAP) is a rare benign adnexal skin tumour of apocrine or apocrine 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
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.

REFERENCES


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

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

*Data not available.

Acta Derm Venereol 88