

## LINEAR MORPHOEA WITH HYPERTRICHOSIS AND ENTRAPMENT NEUROPATHY

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### Summary

A case of linear morphoea on the right thigh of a 24 year old man is described, who presented with hypertrichosis limited to the area of induration and symptoms of entrapment neuropathy in the distribution of medial and intermediate cutaneous nerves of the right thigh. The prolonged use of corticosteroid ointment is attributed as the cause for development of hypertrichosis. The constriction of nerves by sclerosed collagen at sites where the nerves pierced the fascia lata of thigh is put forward as the possible explanation for the symptoms of entrapment neuropathy in this patient.

Morphoea is a collagen disease which usually affects the age group of 20 to 40 years. The lesion develops as a plaque with a waxy yellowish colored centre, a smooth surface and a lilac coloured border. The involved site eventually becomes anhidrotic and alopecic<sup>1</sup>. The elevated plaque may become depressed later. Pigmentation may at times be a conspicuous feature of morphoea and may indeed be the presenting symptom<sup>2</sup>.

The linear form of morphoea is seen predominantly in children. It appears usually as a single unilateral plaque. The usual sites of involvement are the lower extremities, frontal region and thorax. The disease process sometimes extends deep to involve underlying muscle or bone. Frontoparietal lesion may be preceded by bleaching of hair and linear zone of alopecia can occur

on the scalp<sup>3</sup>. Prolongation of sensory chronaxy can occur in morphoea<sup>4</sup>. Segmental morphoea may be associated with arthralgia in the involved extremity<sup>5</sup>. Raynaud's phenomenon has also been reported in cases of circumscribed scleroderma<sup>6</sup>. Other cutaneous abnormalities reported in relation to linear morphoea include warty vascular or pigmented nevi, cafe-au-lait spots, alopecia areata and vitiligo<sup>3</sup>.

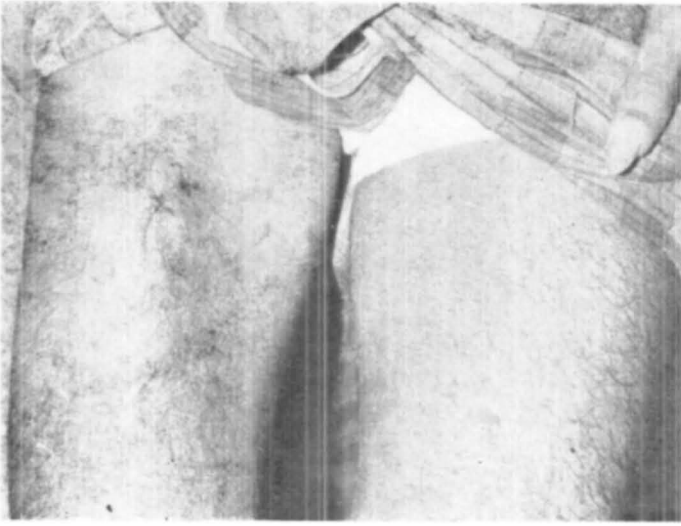
### Case Report

A 24 year old man attended the Dermatology department of Medical College Hospital, Trivandrum on 1st October 1979 for treatment of a linear hirsutic area on the front of his right thigh which was then present for 9 months. One year prior to the presenting problem, patient was diagnosed to have linear morphoea and advised flucortolone (ultralan) ointment application twice daily. After one year of regular application, patient noticed increased growth of coarse, black hairs over the lesion on

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**Fig. 1** Front view of both thighs. Note hypertrichosis in a linear pattern on right thigh.

the thigh. 3 months before his hospital visit, the patient developed numbness and a peculiar sensation of pins and needles under the skin of the antero-medial side of the lower half of his right thigh, extending to the front of of the knee. These symptoms were exacerbated during walking and got relieved on rest. Dermatological examination revealed a linear sclerotic and slightly hyperpigmented  $12 \times 3$  cm sized plaque on the front of the right thigh (Fig. 1). The skin was indurated and bound down. There were numerous coarse black hairs 2 to 3 cm long, seen over the site of induration (Fig. 2). Even a gentle touch over the plaque, evoked severe pain, numbness and tingling sensation on the front and medial side of lower half of thigh and upper part of the knee. The size, power and tone of muscles of thigh were normal. Movements of the knee and hip joints were within normal limits. Except for the sensory disturbances on the thigh there was nothing abnormal in the examination of the nervous system. Other systems were clinically normal. Patient was treated with intra lesional injection of

Kenacort (Triamcinolone acetonide 10 mg/ml) half c. c. once a week for 6 weeks. There was profound relief of neurological symptoms but hairs persisted.

**Investigations**

Routine blood and urine examinations revealed no abnormalities. LF cells were negative. X-ray of chest and thigh revealed no abnormalities.



**Fig. 2** Right thigh of patient—front view. Note coarse black hairs in linear pattern.

Histology of the skin lesion revealed panatrophy of the epidermis.

Basal cell layer showed increased melanin. The dermis was composed of homogenous sclerotic collagen. The subcutaneous fat was replaced by fibrous tissue. The sweat and sebaceous glands were atrophic and the sweat glands were found high in the dermis and bound down by sclerotic collagen bundles. There were cross sections of mature hair follicles about 2 to 3 per low power field. There was sparse mononuclear cell infiltration around the blood vessels. The walls and lumina of blood vessels appeared normal. The hairfollicles were free of inflammatory infiltrate. No nevus cell was seen in epidermis or dermis.

### Discussion

Morphoea is not an uncommon disease. The linear form is predominantly seen in children<sup>1</sup> but our patient is a 24 year old young man. When he was first diagnosed to have morphoea by a dermatologist no hypertrichosis was present. In morphoea dermal appendages generally get atrophied<sup>3,7</sup>.

Pilosity with its several variations has always been and will always be one of the most spectacular manifestations of our speciality<sup>8</sup>. Systemic steroids are well known to cause hirsutism<sup>9,10</sup> and hypertrichosis can result as a complication of topical steroid therapy also<sup>11</sup>. The prolonged use of topical steroid is the only explanation for the development of hypertrichosis limited to the site of morphoea.

Another interesting feature observed in the present case was the symptom of entrapment neuropathy. The medial cutaneous nerve of the thigh divides into an anterior and a posterior branch at the apex of the femoral triangle. Before dividing, the nerve gives off a few filaments which pierce the fascia

lata to supply the skin of medial side of thigh. The anterior branch runs downwards and perforates the fascia lata and supplies the skin as low as the medial side of the knee. The intermediate cutaneous nerve of thigh pierces the fascia lata and divides into 2 branches which descend vertically on the front of thigh and supply the skin down to the knee<sup>12,13</sup>.

Meralgia paresthetica is a condition in which the lateral cutaneous nerve of the thigh gets entrapped at the inguinal ligament and gives rise to paresthesia and pain over the anterolateral aspect of the thigh<sup>14,15</sup>. Meralgia paresthetica can also occur as a result of excess walking, injury, obesity, pelvic tumour, etc. These were not present in our case. Occasionally the nerves may get compressed when they pass through or near rigid anatomical structures. This type of lesion known as entrapment neuropathy involving the medial and intermediate cutaneous nerves of the thigh must have been the cause of the neuritic symptoms in our patient in whom at the area of skin lesion whole subcutaneous fat tissue had been replaced by the newly formed collagen (fibrous tissue) which later got sclerosed. The pathological changes had involved even the muscle tissue. It is possible that the newly formed fibrous tissue is in firm apposition to the fascia lata and fibrous layer of superficial fascia.

It is thus possible to postulate that the neurologic symptoms which this patient experienced are due to entrapment of medial and intermediate cutaneous nerves of thigh and few of their branches at sites where they pierced the fascia lata and fibrous layer of superficial fascia due to pressure or constriction by sclerosed collagen. Though, prolongation of sensory chronaxy has been reported in morphoea<sup>4</sup>, I could not come across any report

of entrapment neuropathy due to morphoea. Fibrosis<sup>16</sup> and thickening of connective tissue<sup>17</sup> have been mentioned among the causes of its aetiology. Intra lesional triamcinolone injection afforded profound relief of neurologic symptoms in our patient. The hypertrichosis persisted. The development of hypertrichosis in this patient cannot be attributed to the associated entrapment neuropathy because, if at all it has any effect on hair growth it is only to produce hypotrichosis rather than hypertrichosis<sup>18</sup>.

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