

Scanning Electron Microscopy in the Diagnosis of Pili Multigemini

Sir,

Pili multigemini is an unusual developmental defect of the hair follicles, in which multiple divided matrices and papillae form multiple hairs contained in a single enlarged pilosebaceous unit (1). These multiple hair shafts emerge through a single pilosebaceous canal as irregular and coarse hairs, thicker than the average. From the first reports by Flemming (2) and Giovannini (3, 4) to the present day, the etiology of this defect has not been clarified.

We describe a new case of pili multigemini and present an ultrastructural analysis of the affected hairs, performed by means of scanning electron microscopy (SEM).

CASE REPORT

An Italian man aged 29 was seen for the presence of "irregular" hairs causing irritation, especially after shaving. Clinical examination revealed several abnormal coarse hairs, 2 to 3 times thicker than the others and dark black in colour, irregularly scattered among normal-looking hair of the beard. The coarse hairs were mainly localized on the chin and on the left side of the bearded areas of the face (Fig. 1). Even if some crusts and erythema at the follicular opening were observed, nodular lesions or true folliculitis were not present. The patients was otherwise healthy and had no history of any skin or general disease of interest. X-ray of clavicles and cranial bones showed no abnormalities. The patients was treated with topic retinoid acid 0.05% and oral vitamin A. No significant improvement was observed after 3 months of treatment.

Permission for histological examination was denied by the patient in fear of an unaesthetic scar. Several abnormal hairs were extracted with a tweezer, fixed in Karnowsky solution, and metallized with gold-palladium for SEM observation.

RESULTS

SEM evaluation of the abnormal hairs revealed multiple linear grooves separated by cords running along the shaft, giving the hair the general aspect of a "bundle of sticks". The abnormal hairs were found to be composed of multiple shafts. These were

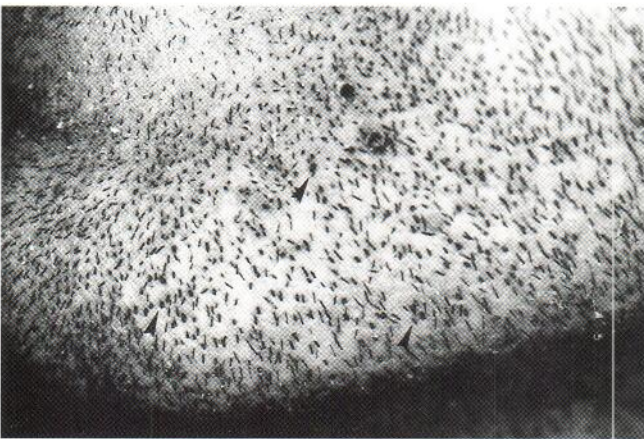


Fig. 1. Left side of the chin: irregularly scattered among normal looking hairs of the beard, several abnormal, coarse, thicker and darker hairs are distinguishable (arrows).

enveloped, compressed and distorted by a single external outer root sheath (Fig. 2), this composition being responsible for the "bundle of sticks" aspect of the hair. Splitting of the shaft was sometimes observed. At higher magnification each branch showed a regular contour of cuticle cells (Fig. 3).

DISCUSSION

Pili multigemini must be suspected in a male patient showing abnormal hairs of the beard, remarkably thicker than the others. Observation with a magnifier may reveal frayed extremities and multiple shafts emerging from a single follicle. Alopecia areata of the beard or inflammatory nodules may be associated features (5, 6), even though no explanation for this association has been proposed. Cleidocranial dysostosis has been reported in one case, but this may have been an accidental combination (6). The abnormal hair can be extracted as a whole, with its outer sheaths. Compared with other reports (5), the extraction of the hair in our patient was not more painful than usual; nor did it require unusual efforts. Histological sections of affected skin areas may show abnormally broad and multigemini hair follicles, with multiple divided dermal papillae (6).

Since it was impossible to perform a skin biopsy, we investigated our patient's abnormal hairs by means of SEM, showing that this is a useful tool in diagnosing pili multigemini. Our SEM investigation of the splittings showed that every dividing branch is coated by its own cuticle, thus ruling out trichoptilosis. These observations are very close to those of Weary's pili bifurcati (7), in which hair shafts split to form parallel branches subsequently rejoining. We believe that pili bifurcati may be a peculiar aspect of pili multigemini.

Pili multigemini must be differentiated from compound hairs, a condition in which the deep parts of the follicles are independent, the hair anomaly being determined by the merging of the follicles in the superficial dermis (5). Tufted hair folliculitis affects the scalp and is characterized by bundles of hairs emerging from a single follicular opening in association with inflammatory changes (8).

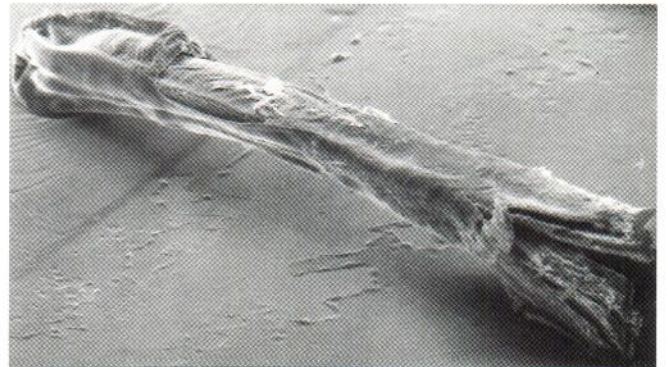


Fig. 2. Three distinct flattened and distorted hair shafts are visible within a common root sheath. The hair shaft shows an irregular profile, resembling a "bundle of sticks" (bar = 1 mm).

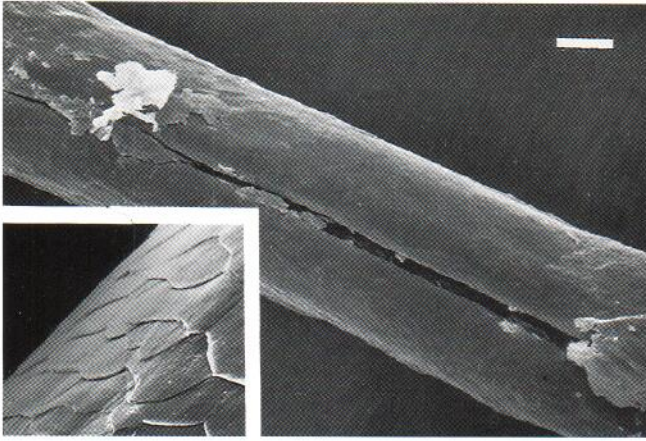


Fig. 3. Splitting occurs along the shaft, without evidence of fracture (bar = 0,1 mm). *Insert*: each branch showed a normal cuticular pattern.

The origin of pili multigemini is still unexplained. Giovannini (3) suggested the reactivation of silent embryonic epithelial germs. Flemming (2) stated that the condition required the merging of different papillae or the subdivision of a single papilla. Pinkus suggested that pili multigemini are determined by the abnormal division of one dermal papilla into multiple papillae (5); multiple papillae produce multiple hair shafts or, when separation between them is not complete, a single thicker and irregular hair shaft reflecting the non-synchronous work of the matrix. More recently, a good deal of attention has been drawn to the bulge area as the source for new follicle generations (9). This area is susceptible to chemical carcinogenesis during the anagen stage. It is possible that mechanical trauma or inflammatory changes may act as stimuli to the proliferation of multiple stem cells inside the bulge, even if it is difficult to

correlate this hypothesis with the rarity of pili multigemini. In the attempt to regulate the proliferation of the epithelial cells of the hair matrix and/or bulge, we treated our patient with topical retinoic acid for 3 months, without any success.

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