Perianal Lentiginosis: A Case of Localized Lentiginosis

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Sir,
Lentiginosis is defined as the presence of lentigines in exceptionally large numbers or in a distinctive distribution. It can be associated with internal diseases but sometimes the lentigines can appear solely as a cutaneous manifestation (1).

CASE REPORT
A 43-year-old woman was seen for asymptomatic multiple perianal pigmented macules of 5 years’ duration and a perianal haemangioma of recent appearance. She had a family history of death by cancer (gastric, pancreatic, uterus and ovarian) and a daughter with a congenital angioma, but no report of family lentiginosis.

On exploration, she presented 23 perianal pigmented macules of 0.3 cm localized outside the demarcation line between mucous skin and normal skin. A red papule of 0.5 cm was found in the left perianal side (Fig. 1). She also had three dermatofibromas on the upper and lower extremities and one epidermal cyst on the chest. No pigmented lesions on nails or mucosas were detected. Her skin was a III skin type of Fitzpatrick classification.

Physical evaluation, electrocardiogram, echocardiogram and radiographic studies of the small bowel with standard barium meal were normal. Studies of the uterus and ovarian echography, cervix cytology, eyes (ophthalmoscopy and tonometry) and audiometry did not show any abnormality. Colonoscopy showed only the angioma of 7 mm diameter in sigmudeum that had been detected at colonoscopy 5 years previously.

Histology of one pigmented macule showed an epidermis with lentiginous configuration with increased pigmentation along the dermo-epidermal junction and with a melanocyte/basal keratinocyte ratio of 3/10. A few melanophages and slight lymphocyte infiltrate were detected in the papillary dermis (Fig. 2). The red papule showed the findings of a capillary haemangioma.

DISCUSSION
The differential diagnosis of lentiginosis (1) includes Peutz-Jeghers syndrome, LEOPARD syndrome and cardiac myxoma syndrome (NAME, LAMB, Carney syndrome). However, no mucosal lesions, intestinal polyps, cardiac myxoma, ocular, pulmonary, genital or hearing involvement were found. Agminated (zosteriform or unilateral) and centrofacial lentiginosis have been published, but our case does not follow the described distributions. Korting has published a case of lentiginosis profusa perigenito-axillary, with lentigines predominantly on axillary and genital areas (2). We could not find any reference to a localized perianal form of lentiginosis.

Perianal lentiginosis can be a separate entity from the lentiginosis already described. The importance of the other cutaneous findings (angiomas, dermatofibromas and cutaneous cysts), or the possibility of association

Fig. 1. Pigmented macules in the perianal region.

Fig. 2. Epidermis with lentiginous configuration with increased pigmentation. Few melanophages in papillary dermis (haematoxylin-eosin, × 80).
with internal malignancy, remains to be determined. Although our case did not show any associated systemic abnormalities, other causes of lentiginosis should be excluded until more cases of perianal lentiginosis have been described. In any case, follow-up seems a reasonable attitude.

REFERENCES