Case Report

Melanocytic nevus associated with syringofibroadenomatous hyperplasia: a case report*

*Nevo melanocítico associado à hiperplasia siringofibroadenomatóide: relato de caso**

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Abstract: The aim of this study was to report a case of a verrucous lesion diagnosed as a compound melanocytic nevus associated with syringofibroadenomatous hyperplasia. The patient was submitted to excision of the abdominal skin lesion and diagnosed as Miesher melanocytic nevus. Optical microscopy revealed epidermal hyperplasia and dermis with nests of nevus cells intimately associated with eccrine duct clusters arranged in a syringoid pattern. The coexistence of epidermal hyperplasia and syringomatoid proliferation is defined as eccrine syringofibroadenomatosis and may result from growth factors released by the impaired stroma.
Keywords: Eccrine glands; Pigmented nevus; Syringoma

***Resumo**: O objetivo deste estudo é relatar um caso de lesão verrucosa cujo diagnóstico foi de nevo melanocítico composto associado à hiperplasia siringofibroadenomatóide. O paciente foi submetido a exérese de lesão cutânea abdominal com diagnóstico clínico de nevo melanocítico de Miesher. A microscopia óptica revelou epiderme hiperplásica e derme com ninhos de células névicas intimamente associados com aglomerados de ductos écrinos e arranjados segundo um padrão siringomatóide. A concomitância de hiperplasia epidérmica e proliferação siringomatóide é definida como siringofibroadenomatose écrina e pode ser devida a fatores de crescimento secretadas pelo estroma alterado.*
Palavras-chave: Glândulas écrinas; Nevo pigmentado; Siringoma

INTRODUCTION

In 1973, Mishima[1] first described nevus cells associated with sweat glands in normal quantities, giving the name *eccrine-centered-nevus* to this finding.

In the case we report this lesion was associated with epidermal and stromal hyperplasia with surrounding clusters of nevomelanocytic cells.

Syringoma associated with basal cell carcinoma was described as an incidental finding in patients undergoing Mohs surgery. The occurrence of melanocytic nevi has been described in association with a variety of benign and malignant conditions such as basal cell carcinoma, trichoepithelioma, pigmented hidradenoma, syringoma and other collision tumors.[2]

The association of nevomelanocytic cells with eccrine ducts is commonly seen in congenital nevi.[1] However, there are no alterations under the epidermis or the dermis or dermal acrosyringeal eccrine or

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ductal proliferation in these lesions. There is eccrine and epidermal duct hyperplasia with stromal fibrosis in the syringofibroadenomatous association.

Eccrine syringofibroadenoma, first described by Mascaro, in 1963, is a benign tumor that occurs mostly in the limbs of elderly patients. Certain immunohistochemical tests suggest acrosyringeal differentiation as the etiology of the lesion, while other studies indicate differentiation around the dermal eccrine duct.[3]

The aim of this paper is to report a case of melanocytic nevus associated with syringomatous hyperplasia in a patient with a verrucous lesion on the abdomen, with a clinical diagnosis of Miescher nevus.

CASE REPORT

A 48-year-old white male patient presented with a Gleason score 3.3 stage $T_{2b}N_0MX$. prostrate carcinoma. Pre-operative examination disclosed a verrucous lesion on the lower abdomen. Clinical examination led to the diagnosis of Miescher nevus, a brownish, rounded verrucous lesion with well-defined borders measuring approximately 1.7 cm along its longitudinal axis. Removal of the lesions was indicated for esthetic reasons.

The patient underwent concomitant radical prostatectomy and excision of the cutaneous lesion in the abdominal region.

The pathological examination described a brown-whitish verrucous lesion with a brownish matte surface and adjacent abdominal region skin measuring 1.7 x 1.2 x 1.0 cm. Microscopy revealed two distinct histopathological features - epidermal and dermal components. The former showed epidermal hyperplasia with delicate anastomotic cords of acrosyringeal epithelium projecting into the papillary dermis, forming fenestrations. The dermal component included grouped nevus cells associated with proliferated eccrine duct clusters both in the papillary and reticular dermis, including altered fibrous and disorganized collagen. The pathological diagnosis was compound melanocytic nevus associated with syringofibroadenomatous hyperplasia.

DISCUSSION

Eccrine ducts, when associated with nevomelanocytic groups, may present in normal numbers, such as in congenital nevi, or in increased numbers, such as in subtypes of syringomas. Morishima et al.[4] analyzed 16 cases of adnexal proliferation associated with melanocytic nevi and classified into three histopathological groups: those associated with hair follicles, those with perifollicular involvement and those centered in eccrine ducts.

In a review of 29 syringoma cases, Patrizi et al,[5] found one case presenting an association of melanocytic nevus cells and syringoma.

Syringomas are rare eccrine benign tumors originated from sweat gland eccrine ducts. They generally develop in the acral region in elderly people. Immunohistochemical testing of this lesion by Mayumi et al. suggested a close association between intraepidermal acrosyringeal ducts and proliferated eccrine ducts within the fibrous stroma, which receive the name syringofibroadenomatous lesion. In another immunohistochemical study, Ohnishi et al.[6] presented similar results to those of Mayumi,[3] and concluded that both sporadic syringofibroadenomatous lesions (also named eccrine syringofibroadenomas[2]) and inflammation induced syringofibroadenomatous hyperplasia[7] had similar morphology.

Eccrine syringofibroadenomas are characterized by epidermal hyperplasia associated with acrosyringeal proliferation, projecting into the papillary dermis where the stroma is fibrous and contains proliferated eccrine ducts. This lesion is more commonly observed in patients with chronic lymphedema, elephantiasis, focal mucinosis, bullous pemphigoid, erosive lichen planus, epithelioid hemangioendothelioma, and surgical incision sites. There seems to be a relation between cutaneous aggression or chronic inflammation and the appearance of eccrine syringofibroadenomas, which in this case would not be a true neoplasm, as the name suggests,[2] but rather a lesion induced by repetitive aggression, which then would be more appropriately named syringofibroadenomatous hyperplasia, as suggested by Mayumi and Ohnishi.

In the case we present, the absence of multiple nevi and syringomas in the same patient means that the random occurrence of a collision between compound melanocytic nevi and syringomas or eccrine syringofibroadenomas,[8] is improbable. It is important to mention it is an association, that is, there are nevic cells surrounding the proliferated eccrine duct walls (Figures 1 and 2), unlike a collision lesion, where these characteristics are anatomically very close and have no etiological or pathological relation with congenital nevus and syringomas.[8,9]

The simultaneous occurrence of melanocytic nevi and adnexal lesions takes place in benign and/or malignant conditions including trichoepithelioma, pigmented hidradenoma, basal cell carcinoma and syringoma, most of which are collision lesions.[2,7]

Imagawa et al.[9] studied the recurrence mechanism of nevi following dermabrasion procedures, and demonstrated the close relationship between nevic cell proliferation and sweat gland eccrine ducts. Based on the pathophysiology of the recurrence

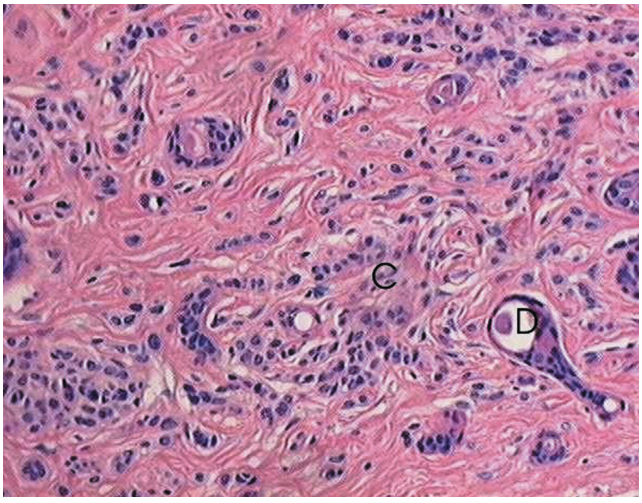

FIGURE 1: Clusters of intradermal nevic cells (C) associated with eccrine ducts (D)

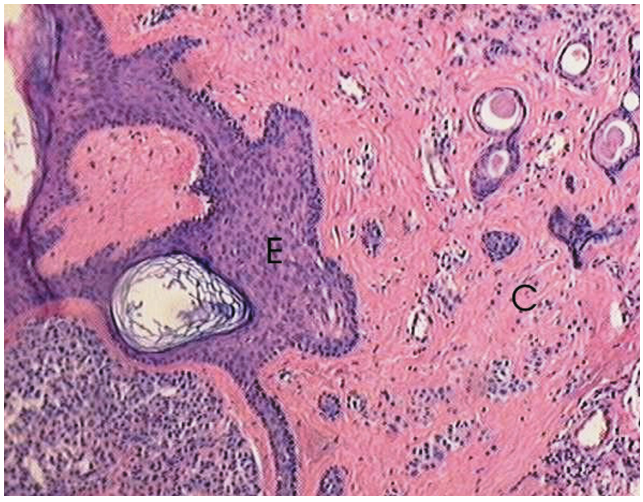

FIGURE 2: Epidermal hyperplasia (E) with thin anastomotic cords of acrosyringeal epithelium forming fenestrations into the dermis (C)

mechanism of melanocytic nevi, it has been suggested that nevic cells originate from the acrosyringeal cells present in the dermal epidermal junction and in papillary dermis eccrine ducts.

Mishima et al.[1] discussed the origin of the nevosyringomatous association lesion, raising two possibilities. The first one assumes that nevoblasts are part of sweat gland duct walls; therefore, when they proliferate due to some stimulus, eccrine ducts also are stimulated and proliferate. The second theory suggests a possible syringotropism at the nevic cell proliferation site, that is, the nevic growth or proliferation factor would cause acrosyringeal tropism and duct cell proliferation.

In a literature review, generally speaking growth factors are possibly the main stimuli inducing proliferation and dysplasia in the site they operate. Inflammation due to repetitive trauma or local inflammatory disease would then, according to this theory, trigger syringomatous proliferation.[2,6]

In this case the patient had no local aggression factor. No association between this type of lesions and prostrate carcinoma was found in literature. Therefore our case appears to be a sporadic case of an association lesion of a compound melanocytic nevus and syringofibroadenomatous hyperplasia. ❑

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