Melanoma *in situ* and syringoma: a rare collision tumor. Clinical-pathological report of a case*

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Abstract: Collision or contiguous tumors, defined as two or more distinct tumors occurring at one site, are often an unexpected finding and may represent a diagnostic challenge, as clinical and histological presentations do not always coincide. Various combinations of collision tumors have been described with respect to melanocytic lesions, with the most frequently reported being the combination of nevus and basal cell carcinoma. We present an unusual case on the nose involving a melanoma *in situ* and a clinically-inapparent syringoma, which, to the best of our knowledge, is the first report of this combination.
Keywords: Melanoma; Immunohistochemistry; Lentigo; Syringoma

INTRODUCTION

Cutaneous neoplasms with two or more distinct cell populations are rare but well documented, and may pose a diagnostic challenge to clinicians and pathologists. They often mimic other cutaneous tumors or present as nonspecific neoplasms. Diagnosis of a collision tumor may be an accidental and unexpected histological finding. Many combinations of collision tumors have been described, with the most frequently reported being the combination of nevus and basal cell carcinoma (BCC). A melanocytic tumor in collision with a benign or malignant epithelial neoplasm is less commonly documented. Some collision tumors involve melanomas, most frequently in combination with a BCC. [1-3] Even more exceptional is the involvement of an adnexal tumor in a collision tumor. Syringomas are relatively common benign tumors that originate in the sweat glands. They present clinically as 1 to 5 mm yellowish, rounded or flat-topped dermal papules. They are generally multiple, and are distributed symmetrically on the chest, neck, or face, and, especially, the lower eyelids.[4] We report an unusual case of a collision tumor on the nose involving a melanoma *in situ* and a syringoma. To the best of our knowledge, this is the first report of this combination.

CASE REPORT

An 82-year-old Caucasian man with a history of atopic dermatitis and osteoarthritis presented with a brown-colored asymptomatic macular lesion on the left nasal root. The lesion had grown progressively for approximately two years and was untreated. Examination revealed an ill-defined 15 mm x 7 mm macule with an irregular shape and asymmetrical distribution of the pigment network (Figure 1). The clinical impression was of lentigo maligna and a punch biopsy was performed to confirm the diagnosis.

Microscopic study of the biopsy revealed lentiginous melanocytic dysplasia, which involved the hair follicles and was positive for Melan-A, HMB45 and S-100 stains. Subsequently, the lesion was completely excised by surgery. Histopathologically, the surgical specimen revealed two distinct histological components. In the basal layer, there were melanocytic dysplasia and nucleocytoplasmic abnormalities with an irregular distribution and involving hair follicles. In the upper and middle dermis there were clusters of small ducts, occasionally with comma-shaped extensions, lined by a double-layered epithelium typical of a syringoma (Figure 2). Immunohistochemical staining confirmed that melanoma was positive

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for Melan-A and HMB45 and that syringoma was positive for the carcinoembryonic antigen (CEA) (Figure 3).

DISCUSSION

In recent years, various terms have been used to describe two lesions occurring in one site, resulting in increasing terminological confusion. Satter *et al.* proposed a classification to simplify the nomenclature of these lesions, classifying them as collision, combined, colonized, or biphenotypic tumors.[5] Collision tumors are defined as two distinct neoplasms that occur within close proximity of each other but maintain sharply distinct boundaries. Neoplasms consisting of two phenotypically different, yet imperceptibly intertwined populations of malignant cells are referred to as combined tumors, and immunohistochemical stains are often required to appreciate the two tumor cell populations. Colonization describes a situation where large dendritic melanocytes populate another neoplasm, e.g., a melanoma *in situ* colonizing a BCC. Biphenotypic tumors are exceptionally rare neoplasms that arise from a common stem cell precursor that undergoes divergent differentiation.

In our patient, the syringoma was not clinically apparent and there were no syringomas in other facial areas. Melanoma and syringoma were close but separated, indicating a collision tumor; this impression was supported by the immunohistochemical data, which clearly differed and did not overlap. Immunostaining with CEA revealed an epithelial syringomatous population, while HMB45 positivity in the melanocytic population confirmed the presence of a melanoma *in situ*, thus drawing a clear boundary between the two tumors. This, together with the fact that the initial biopsy only showed the melanocytic component and not the syringoma, confirmed that this was a collision tumor.

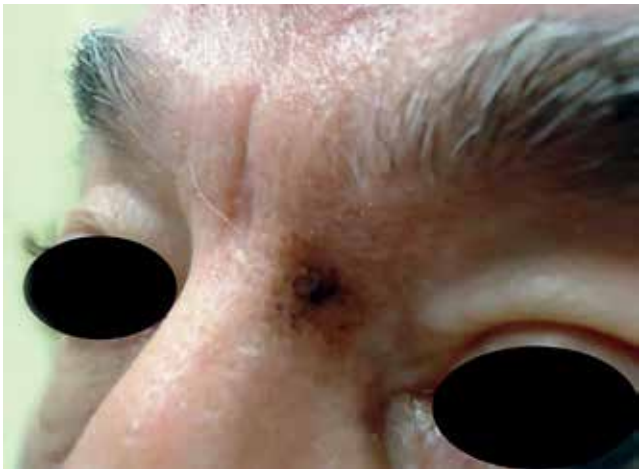

Figure 1: Irregular, asymmetric pigmented lesion on the left nasal root

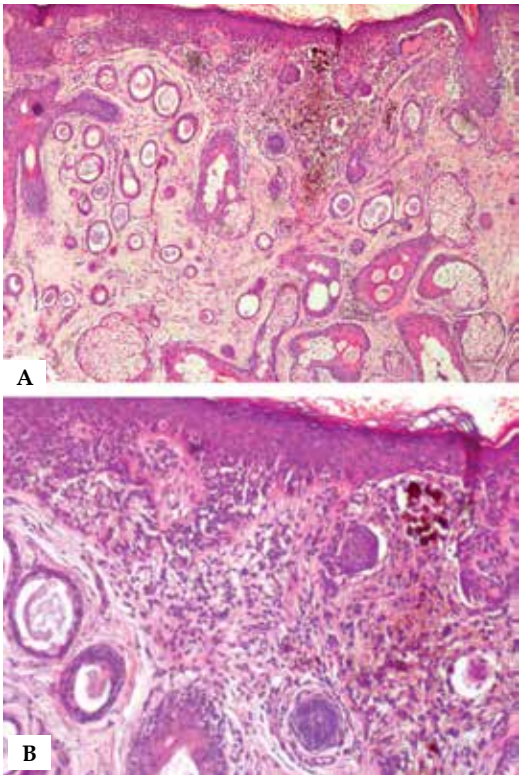

Figure 2: A: Hematoxylin & eosin staining of melanoma *in situ* with follicular involvement and syringoma structures in the middle dermis (X2). **B:** Detail of the collision between the melanoma in situ and the syringoma (X200)

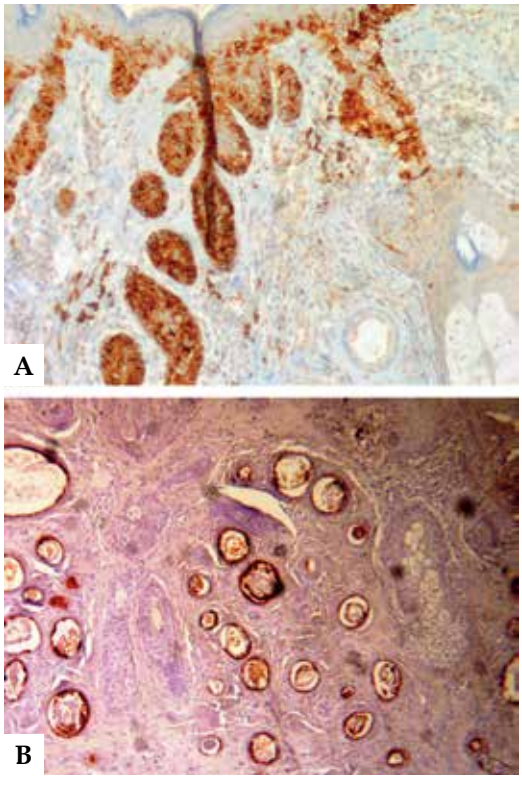

Figure 3: A: Melan-A staining showing the melanoma *in situ* with follicular involvement (x200). Duct structures do not stain. **B:** CEA staining showing the ducts of the syringoma (X100)

Reactive eccrine gland ductal proliferation has been reported in a variety of inflammatory skin diseases (e.g. scarring alopecia) and benign and malignant neoplasms. Guitart *et al.* proposed the term of 'syringomatous dermatitis' for those cases of reactive hyperplastic response of the eccrine duct resulting from an inflammatory skin reaction.[6] In our case we found sclerotic dermal changes in the area of the syringoma, the ducts were located in the middle dermis and some presented with typical comma-shaped structures. These findings, along with the absence of a previous inflammatory cutaneous process, are distinguishable from reactive eccrine gland ductal proliferation and confirm the diagnosis of a syringoma.

To date, a few cases of collision tumors involving a syringoma have been reported in association with intradermal nevi, BCCs, epidermal cyst and Spitz nevus.[3,4,7-10] Whether the coexistence of a syringoma with these conditions occurs by chance alone remains unclear, but it may be that the silent form of this tumor occurs more frequently than suspected.[4] Syringomas are common lesions found on the face, and therefore may occasionally be found in biopsies or excisions of facial lesions performed for unrelated pathologies, just like solar lentigines, seborrheic keratosis or actinic keratosis.

We report the first case of a collision tumor involving a syringoma and a melanoma *in situ*. Syringomas are common, but their association with other types of tumor is rare. Histopathology and immunohistochemical staining were very helpful in distinguishing between the two components. Complete excision biopsies are necessary to establish the diagnosis of collision tumors, and long-term follow up is recommended.❑

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