Title
A unexpected growth arising within nevus sebaceous of Jadassohn

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Abstract

The predisposition to epithelial neoplasms in nevus sebaceous is well established; most tumors occur in adults and are benign. Hidradenoma is a relatively rare benign tumor of sweat gland origin that can rarely arise within a nevus sebaceous. We present an interesting case of a hidradenoma and sebaceoma arising within a nevus sebaceous and present a literature review of the 2 conditions. Even though hidradenoma is a benign tumor, we would advocate complete excision given the potential for malignant transformation.

Case synopsis

A 53-year-old woman presented with a 6-year history of an enlarging pink soft nodule arising within a congenital lesion on the left temple. She had a biopsy two years previously, but declined treatment on the basis that it was a benign growth. Clinical examination revealed a dark pink nodule measuring 18mm x 15mm on the left temple arising within a light brown warty plaque (Figure 1). On the lateral edge there was an additional smaller pink crusted papule measuring 5mm x 6mm. The whole lesion was excised with clear margins and the defect repaired with a skin graft.

Figure 1. Dark pink 18 x 15mm nodule arising within a pale plaque with a smaller pink papule at the inferior edge
Histological Findings

Excisional biopsy of the pink nodule (left superior-medial) shows a well-defined lobulated lesion comprising ductular structures and sheets of squamoid cells showing no atypia. An eosinophilic hyalinized stroma forms the tumor matrix. There are occasional cells showing intra-cytoplasmic vacuolation (Figures. 2a-2d).

The adjacent skin includes another well-defined lesion involving epidermis and superficial dermis comprising islands of basaloid epithelial cells around foci of mature sebocytes (Figures. 3a-3b). Occasional mitotic figures are seen.

Figure 2a: Well defined lobulated lesion showing cystic and solid areas of the lesion  
Figure 2b: Ductal structures within hidradenoma  
Figure 2c: Higher magnification of hidradenoma showing squamoid differentiation and intracytoplasmic lumina  
Figure 2d: Focal sebaceous differentiation within hidradenoma  
Figure 3a: Sebaceoma in adjacent skin  
Figure 3b: Higher magnification of the sebaceoma

Diagnosis: Hidradenoma and sebaceoma arising within a nevus sebaceous.

Discussion
Nevus sebaceous of Jadasson (NS) is a well-demarcated hamartomatous plaque mainly composed of sebaceous glands. The most commonly affected area is the scalp, but they can also occur on the face, neck, and trunk. The appearance generally manifests as a solitary tan or orange-yellow smooth plaque, which may have a warty or waxy texture. It can become more pronounced after puberty.

The predisposition to formation of certain epithelial neoplasms in NS is well established[1]. The most common neoplasms reported to arise are benign neoplasms such as trichoblastoma syringocystadenoma papilliferum (SP), and trichoolemmoma, followed by malignant neoplasms such as basal cell carcinoma (BCC), sebaceous carcinoma, and squamous cell carcinoma[2]. In a study of 596 cases, benign tumors occurred in 13.6 % of adults compared with 1.7% of children. Historically it has been reported that rates of BCC occurring in NS were as high as 10-20%. However, this has now been clarified to be less than 1%[2]. The discrepancy in percentages may have been related to histologic misinterpretation of benign trichoblastomas and basal cell hyperplasia as BCC[2].

Hidradenoma is a relatively rare tumor of sweat gland origin and was first described by Liu et al in 1949. Hidradenoma usually presents as a solitary dermal nodule, although it can be cystic and highly vascularized. It is more common in females, and can be found on sites of the body with a predilection for the middle and older age groups. Hidradenoma arising in NS is rare and only 8 cases have previously been reported[3]. Malignant hidradenoma is also rare with less than 50 cases ever reported in the literature. It is aggressive with local recurrences reported in more than 50% of surgically treated cases and usually develops de novo. However, a handful of cases showing malignant transformation from its benign counterpart have been reported[4,5].

Sebaceoma presents as a yellowish to skin-colored papule or nodule and has been reported to arise within NS[6]. A combination of hidradenoma and sebaceoma arising within NS has not been documented yet in the literature.

Although hidradenoma is a benign neoplasm and very rarely arises in NS, it is important that it is correctly identified and treated because it has the potential for malignant transformation[4].

References


LEARNING POINTS

1. The predisposition to epithelial neoplasms in nevus sebaceous is well established; most tumours occur in adults and are benign.
2. Hidradenoma is a relatively rare benign tumour of sweat gland origin that can rarely arise within a nevus sebaceous.
3. Even though hidradenoma is a benign tumour, we would advocate complete excision given the potential for malignant transformation.