Spiradenocylindroma with Trichoepithelioma – A Collision Tumor with Multiple Differentiation

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Abstract: Spiradenoma & cylindroma are relatively uncommon sweat gland tumors. These lesions along with trichoepithelioma can co-exist and this gives support to the theory that they are part of the same spectrum and have a similar derivation. They can occur as separate lesions or within a single tumor mass as collision tumors. Here we report a case of a 67 year old female who presented with multiple papulonodular lesions over the nose for 10 years, which was clinically suspected to be an adnexal tumor. Biopsy of two different lesions revealed a spiradenocylindroma & trichoepithelioma.

Keywords: Benign adnexal gland tumors, collision tumor, eccrine and apocrine differentiation, Spiradenocylindroma, Trichoepithelioma

1. Introduction

Tumors of sweat glands are uncommon with a wide histological spectrum and complex classification. Many eccrine/apocrine lesions coexist with composite differentiation. Eccrine sweat glands are present almost everywhere in the skin while apocrine sweat glands are found mainly in the axillae, groin and anogenital regions. Many tumors assumed to have an eccrine origin are now recognized to have apocrine counterparts as well.While the phenotypic features differ between cylindromas and spiradenomas, recent studies have shown immunohistological and cytomorphological overlap, with both tumors exhibiting apocrine, eccrine, secretory and ductal features^[1]. The coexistence of more than one cutaneous adnexal neoplasm in a single biopsy specimen is unusual and is most frequently recognized in the context of a nevus sebaceous or Brooke-Spiegler syndrome, an autosomal-dominant inherited disease characterized by cutaneous adnexal neoplasms, most commonly cylindromas, spiradenomas and trichoepitheliomas^[2].Collision tumors refers to the presence of two tumor types in a single lesion^[3]. In spiradenocylindroma, features of both tumors co-exist, further establishing that they can have a common origin^[4].

2. Case History

A 67 year old female presented to the skin OPD with complaints of raised skin lesions over the nose for the past 10 years. There was no history of pain or similar lesions in the family. On examination, multiple erythematous nodules with dome shaped appearance were present measuring 4x3x1 cms and the largest papule over the proximal nasal bridge measured 1x1 cm[Plate 1]. Clinically it was suspected to be a benign adnexal tumor. Biopsy of a nodule and a papule was taken [Plate 2].

3. Histopathology

Skin biopsy of the nodule showed two different patterns of tumor [Plate 3] – one is closely packed with deeply basophilic cells arranged as a sheet with circumscription and surrounded by dermal collagen. The appearance is consistent with the diagnosis of spiradenoma {Fig.1, 2}.

The other lesion is composed of numerous islands of epithelial cells of varying size and shape. The islands are surrounded by hyaline sheath and a narrow band of collagen. The islands of epithelial cells fits together as pieces of a jigsaw puzzle and have mosaic like masses. Two types of cells are seen in this tumor – cells with small, dark staining nuclei are seen in the periphery and large cells with a light staining nucleus are present in the center. This appearance is consistent with the diagnosis of cylindroma {Fig.1, 2, 3, 4}.

Since the characteristic histological pattern of both tumors namely spiradenoma & cylindroma are seen in the same section, the diagnosis of spiradenocylindroma has been made.

Skin biopsy of the papule [Plate 4] also showed different patterns of lesions namely, spiradenoma, which was located in the dermis as lobules without any connection to the epidermis. The lobules are evenly and sharply demarcated which issurrounded by a fibrous capsule. Large pale cells are grouped around the lumina and smaller darker cells are found in the periphery {Fig.5, 6}.

The second lesion was well circumscribed, small & symmetric in nature and was identified as trichoepithelioma. Collection of hyperchromatic epithelial cells (primitive epithelial buds) with circumscribed spaces between them, containing abortive hair shafts and cystic spaces filled with proteinaceous material was seen {Fig.7, 8, 9, 10}.

4. Discussion

Skin adnexal tumors (SAT) are derived from primordial germ cells which can exhibit varying morphological differentiation towards one of the different types of adnexal epithelium present in normal skin i.e., follicular, sebaceous, eccrine or apocrine. The diagnosis of mixed SAT relies on histological evaluation and they are usually classified according to the predominant morphological component^[5]. The histogenesis of mixed adnexal tumors is still uncertain; however the possibility of origin from pluripotent stem cells can be considered. The pluripotent cells may differentiate towards more than one type of appendage giving rise to a tumor that contains elements of two or more appendages in varying degrees of maturation^[6]. This has been observed in our case, where areas of eccrine, apocrine and follicular differentiation has been observed. In addition, the coincidence of spiradenoma and cylindroma with trichoepitheliomas reported in the past support the notion that both neoplasms derive from pluripotential cells of the abortive follicular-apocrinesebaceous 'anlage'^[7]. Moreover these lesions have also occurred in a sporadic fashion in an older individual which is uncommon.

5. Conclusion

Skin adnexal neoplasms always require histopathological examination and collision tumor is one such histopathological diagnosis.

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PLATE 1 : Clinical Picture



<u>PLATE 2</u> : Post biopsy picture



PLATE 3 : SPIRADENOCYLINDROMA



Figure 1: H&E Stained 4x

Tumor is seen in the dermal region. Thin arrow indicates Spiradenoma composed of closely packed hyperchromaticcells. Thick arrow indicates Cylindroma, arranged as islands & surrounded by hyaline sheath



Figure 2: H&E Stained 10x

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Figure 3: H&E Stained 10x Tumor cells fit together as pieces of jig-saw puzzle.



Figure 4: H&E Stained 40x Dark staining hyperchromatic cells seen in the periphery & lighter staining cells are seen in the center.

PLATE 4 : SPIRADENOMA WITH TRICHOEPITHELIOMA



Figure 5: H&E Stained 4x Sharply demarcated lobules of Spiradenoma



Figure 6: H&E Stained 10x



Figure 7: H&E Stained 4x A cyst lined by benign flattened squamous cells & filled with lamellatedproteinaceous material is seen.

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Figure 8: H&E Stained 10x



Figure 9: H&E Stained 4x Spiradenoma with Trichoepithelioma



Figure 10: H&E Stained 10x Abortive hair shaft surrounded by epithelial cells