Spiradenocylindroma of Skin: A Hybrid Tumor

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Abstract

Spiradenocylindroma is a rare occurrence where tumor has histological features of both spiradenoma and cylindroma in single nodular lesion. Due to dual histomorphological features in a single lesion, it is considered as a hybrid tumor. We describe a case of spiradenocylindroma in a 70-year old female. Spiradenocylindroma presents a diagnostic challenge to the histopathologist because both tumors share some common features; however, the presence and absence of lymphocytes are important for differentiation.

Keywords: Cylindroma, histomorphology, hybrid, spiradenoma

Introduction

Spiradenoma and dermal cylindroma are benign cutaneous neoplasms usually presenting as solitary or multiple nodular lesions. These two tumors may present as separate nodules or may have features of both tumors in a single lesion. The word “spiradenocylindroma” is specifically reserved for the lesions which have histological features of both spiradenoma and cylindroma in the same tumor nodules. The most frequent locations for these tumors are scalp and face followed by head and neck region. Here, we document a case of spiradenocylindroma occurring in the preauricular region of an old female patient.

Case Report

A 70-year-old female patient presented with a slow-growing swelling on the left side of the face for the past 3 years. Physical examination revealed a nodular subcutaneous mass measuring 3 cm × 2.5 cm in the left preauricular region, and clinical impression of adnexal tumor was suggested. Fine-needle aspiration cytology of the swelling yielded hemorrhagic material. The smears were cellular showing multiple clusters and discrete small, round to oval cells with hyperchromatic nuclei, inconspicuous nucleoli, and scanty cytoplasm. Background showed deposits of dark magenta-colored pinkish globular basement membrane type material [Figure 1]. The globules were occasionally surrounded by basal type of cells. Occasional cells with elongated nuclei were also noted. A diagnosis of benign adnexal tumor, likely cylindroma, was offered. Total resection was performed with 1 cm margins. Grossly, a skin covered nodular lesion was identified measuring 4 cm × 3 cm × 2.5 cm. Cut surface showed a relatively circumscribed tumor measuring 3 cm × 2.5 cm × 1.5 cm, with gray-white to gray-tan areas [Figure 2a-c]. Histopathologically, two distinct tumor areas were observed. One area had typical spiradenoma pattern, and other had typical cylindroma pattern [Figure 3]. The spiradenomatous part showed a well-circumscribed, tumor mass having two types of neoplastic cells: dark and pale which were surrounded by a thin basement membrane. The pale cells were arranged in discrete aggregates within the nodules. A distinctive feature of lymphoid infiltrate infiltrate around the blood vessels and within the neoplastic nodules was also observed [Figure 4]. Cylindromatous area was composed of small tumor cell aggregates that were distinctively rectangular, triangular, and polyhedral in shape and arranged in a jigsaw puzzle pattern [Figure 5]. These tumor cell aggregates were surrounded by thick, homogeneous eosinophilic basal membrane material. The neoplastic cells of these areas consisted of cells having two different appearances: (i) peripheral cells aligned in a palisade with small dark nuclei and with a small amount of cytoplasm and (ii) the remaining cells with nuclei that were...
Figure 1: Fine-needle aspiration smear shows clusters and discrete small, round to oval cells with magenta-colored basement membrane material (MGG, ×200)

Figure 2: (a) Preauricular swelling. (b) Grossly, externally nodular. (c) Cut surface is gray-white with focal gray-tan and firm to hard tumor

Figure 3: Photomicrograph shows features of both spiradenoma and cylindroma tumor patterns (H and E, ×100)

Figure 4: Photomicrograph depicting spiradenomatous part of tumor with lymphoid infiltrate within the neoplastic nodules (H and E, ×400)

larger, plumper, and have more copious, paler cytoplasm. The lymphoid cells within the cylindromatous component were scanty. Cylindromatous and spiradenomatous areas were often intimately intermingled; however, cylindromatous areas were predominantly seen in our case, and there was no well-defined transition zone.

Discussion

Spiradenocylindroma is a rare occurrence where tumor has features of both spiradenoma and cylindroma in the single nodular lesion.\(^1\)\(^2\) It is more common in females and usually present with single or multiple nodular lesions of variable size, especially on scalp and face.\(^1\) Due to dual histomorphological features in a single lesion, it is considered as a hybrid tumor.\(^3\) Histologically, the cells of spiradenoma form large, well-circumscribed lobules with two distinct types of epithelial cells. One cell type contains small, hyperchromatic nuclei and the second contains larger, pale nuclei, which are frequently arranged around a lumen to form a duct-like structure.\(^4\) Lymphocytic infiltration is characteristic of spiradenomas and is helpful in distinguishing spiradenomas from cylindromas. The tumor is composed of two cell types, similar to those of spiradenoma, rimmed by prominent periodic acid–Schiff-positive basement membrane material.\(^4\)\(^5\) In addition, this basement membrane material is seen within tumor cells as hyaline droplets.\(^2\) Nests of irregularly shaped tumor islands arranged adjacent to one another have classically been described as reminiscent of a jigsaw puzzle pattern. By contrast with spiradenoma, cylindroma has few or no lymphocytes.\(^2\)\(^4\)\(^5\)

The two tumor components, spiradenoma and cylindroma, have some similarities and some distinctive features as well. Cylindromas and spiradenomas share common features such as the presence of homogeneous eosinophilic globules within the cellular aggregates and prominent tubules that sometimes show signs of apocrine changes in such aggregations.\(^6\) Cylindromas, unlike spiradenomas, show the deposition of abundant basement membrane-like material in between the tumor cells.\(^7\) Spiradenoma further differs from cylindromas by the focal occurrence of elongated tubules, lymphoid infiltrate,
squamous differentiation, widely gaping vessels with thrombi, and perivascular fibrinoid material.\(^6\)

The histochemical and immunohistochemical studies have not clarified the histogenesis of spiradenoma.\(^6\) The frequent association of spiradenoma and cylindroma, a likely apocrine neoplasm, and the sporadic association of spiradenoma with neoplasms of follicular differentiation such as trichoepithelioma support an apocrine line of differentiation for spiradenoma on the basis of common embryologic origin for the three elements of the folliculosebaceous-apocrine unit.\(^8\) This is furthermore supported by some examples of spiradenoma that show decapitation secretions in the cells lining the luminal border of the tubular structures.\(^6,8\) Therefore, the qualifying term of eccrine that almost invariably is applied to spiradenoma is inaccurate.\(^2\)

Immunohistochemically, all lymphoid cells within the tumors react positively with antibody to leukocyte common antigen and different proportions of these cells react with CD20 and CD45R antibodies, therefore proving their mixed B- and T-cell nature.\(^5,6\)

The role of lymphoid tissue in spiradenomas and their scarcity or absence in cylindromas is difficult to explain. The selective presence of lymphocytes in spiradenoma and their absence in cylindroma suggests that spiradenomas have unique property of attracting lymphocytes.\(^4,9\) Prognosis of cylindroma and spiradenoma is good; however, malignant transformation is uncommon but spiradenomas have high propensity to malignancy as compared to cylindromas. Simple excision is usually curative for both tumors.\(^1,10\)

The term “spiradenocylindrocacinoma” is proposed for the malignant counterpart.\(^9\) Clinical features of malignant change include ulceration, rapid growth, pain, and change in color. Thus, tumors showing rapid enlargement should undergo a careful pathological examination for a possible malignant transformation.\(^5\)

**Conclusion**

Spiradenocylindroma presents a diagnostic challenge to the histopathologist because both tumors share some common features; however, presence and absence of lymphocytes is important for differentiation. Histochemistry and immunohistochemistry have limited role in diagnosis.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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Figure 5: Photomicrograph shows jigsaw puzzle pattern of cylindromatous part (H and E, ×100)