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Research Article

Clinicopathological Characteristics of 11 Cases with Spiradenoma: A Rare Adnexal Tumor

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Abstract

Objective: To explore the clinicopathological characteristics of spiradenoma (SA), and make exact diagnosis and differential diagnosis of this disease. Methods: The clinical and pathological data of 11 patients with spiradenoma in the First Affiliated Hospital of Jinan University from 2013 to 2022 were retrospectively analyzed, and combined with the review of literatures. Results: Among the 11 patients with spiroadenomas, 6 cases were male and 5 were female; the age ranged from 19 to 70 years old, with an average age of 41 years; Spiradenomas are more common in the trunk and limbs. At low-power microscope, the tumor mass with capsule in the dermis. Under a high-power microscope, the tumor is usually composed of two types of cells, which are the marginal small basaloid cells with dark staining and central larger cells with a pale and acidophilic nucleus. Other morphological structures included 2 cases of cystic solid and cylindroma respectively. Immunohistochemistry showed that epithelial and myoepithelial differentiation. Conclusion: Spiradenoma often occurs subcutaneously and requires histopathological diagnosis to avoid misdiagnosis. Its biological behavior is benign and its prognosis is good. But malignant transformation should be considered in some case with long course or relapses. SA is rare in clinical and is prone to misdiagnosis or missed diagnosis. This study summarizes the clinilcopathological characteristics of SA, providing reference for future clinical diagnosis and treatment.

Keywords

Spiradenoma, Pathological Features, Differential Diagnosis

1. Introduction

Spiradenoma (SA) is a rare benign tumor of the skin adnexal, which was first described by Kerting and Helwig in 1956 [1]. In the past, spiradenoma were believed to be tumors originating from eccrine, but existing evidence suggests that spiradenoma can differentiate into apocrine gland [2]. SA tends to be solitary, but when multiple rashes are observed, we should consider with Brooke-Spiegler syndrome (BSS) [3]. Which is an important clue to confirm certain genetic diseases. Due to lack of characteristic clinical manifestations, it is easy to be misdiagnosed. Although SA is rare and mostly benign, it can occasionally transform into malignancy. Spiradenocarcinoma is a rare skin malignant tumor first reported by Dabska et al. in 1972 [4]. It is often caused by long-term asymptomatic malignant transformation of benign SA [5]. Currently, only a few cases have been reported. This article analyzes and summarizes the clinical and pathological

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characteristics of SA and spiradenocarcinoma, aiming to improve understanding of this disease, reduce misdiagnosis rates, and provide reference for future clinical diagnosis.

2. Materials and Methods

2.1. Materials

Collect data from 11 patients diagnosed with SA at the First Affiliated Hospital of Jinan University from August 2013 to March 2022. After review by two pathologists, there were 11 cases of benign SA.

2.2. Methods

Formalin-fixed (10%), air-dried smearsand and stained with HE. Retrospective analysis of clinical data (age, gender, lesion location, characteristics, etc.) and histopathological features.

3. Results

3.1. Clinical Characteristics

Among the 11 cases of SA, there were 6 males and 5 females, Age ranged from 19 to 70 years old, with an average age of 41 years old. The course of the disease ranged from six months to 10 years; 2 cases occurred in the head and neck, 5 cases in the trunk, and 4 cases in the limbs. Among the 11 cases of benign diseases, one 41 year old male patient developed a new nodule at the old scar with mild tenderness one year after lipoma resection. In this group of cases, the initial diagnosis was one case of lipoma, 3 cases of epidermal cyst, and 7 cases of skin tumor.

3.2. Clinical Features

The main manifestation of lesion is intradermal nodules or papule, with a diameter of mostly 0.8-2.0cm, and the color is normal skin color, light pink, or blue. (Figure 1a-1b).

3.3. Pathological Features

Under low-power microscope, 11 cases of SA mostly located in the dermis and also visible in the fat layer. The tumor was surrounded by a fibrous capsule, with clear boundaries (Figure 2a). Under a high-power microscope, 11 cases showed typical histological features, consisting of two types of cells. Surrounded by small cells with dark nucleus and larger cells with pale nucleus in the center (Figure 2b). The mixture of the two forms a large consolidation, some tumor cells were arranged in a cribriform and rosette-like pattern. The background showed few scattered lymphocytes, and occasionally visible eosinophils. Normal blood vessels and sweat glands are often distributed at the edge of the tumor capsule. 7 cases showed obvious ductal differentiation and glandular cavity formation in focal lesions, with eosinophilic amorphous material in the cavity (Figure 2c). Two cases showed vascular lumens filled with red blood cells in the local area (Figure 2d). Two cases were cystic, with dilated lymphoid vessels, pink amorphous stromal matrix material could be seen. The other two cases of coexisting cylindroma, some of which exhibit histological features of SA, while others are composed of tumor masses of varying sizes like “jigsaw”, surrounded by eosinophilic basement membrane--like materia and lymphocytes are absent.

3.4. Immunohistochemical

EMA marked central larger cells which belong to sweat gland lumen structure, SMA and P63 marked surrounding small cell which differentiation into myoepithelial cells (Figure 3a-3c).

3.5. Treatment and Prognosis

Surgical resection after local anesthesia, three patients were lost to follow-up, and eight patients have been followed up to date. Currently, the prognosis is good and there is no recurrence.

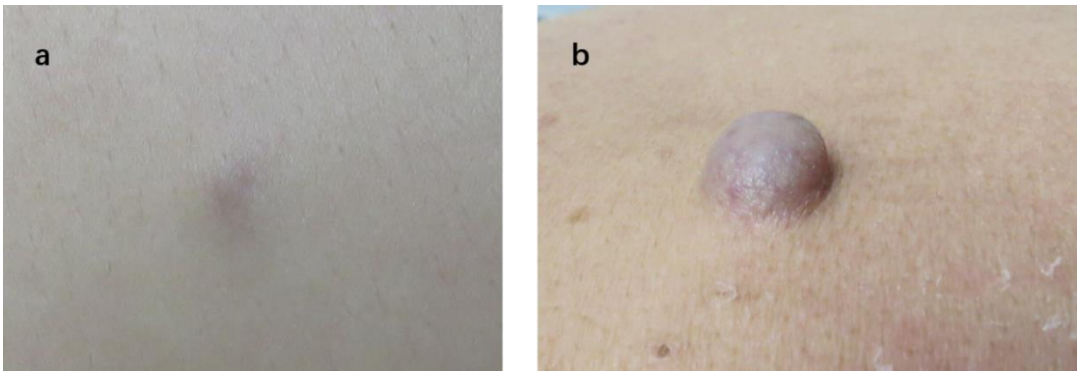


Figure 1. *(a-b): Clinical manifestations of SA: normal skin color and bule papules.*

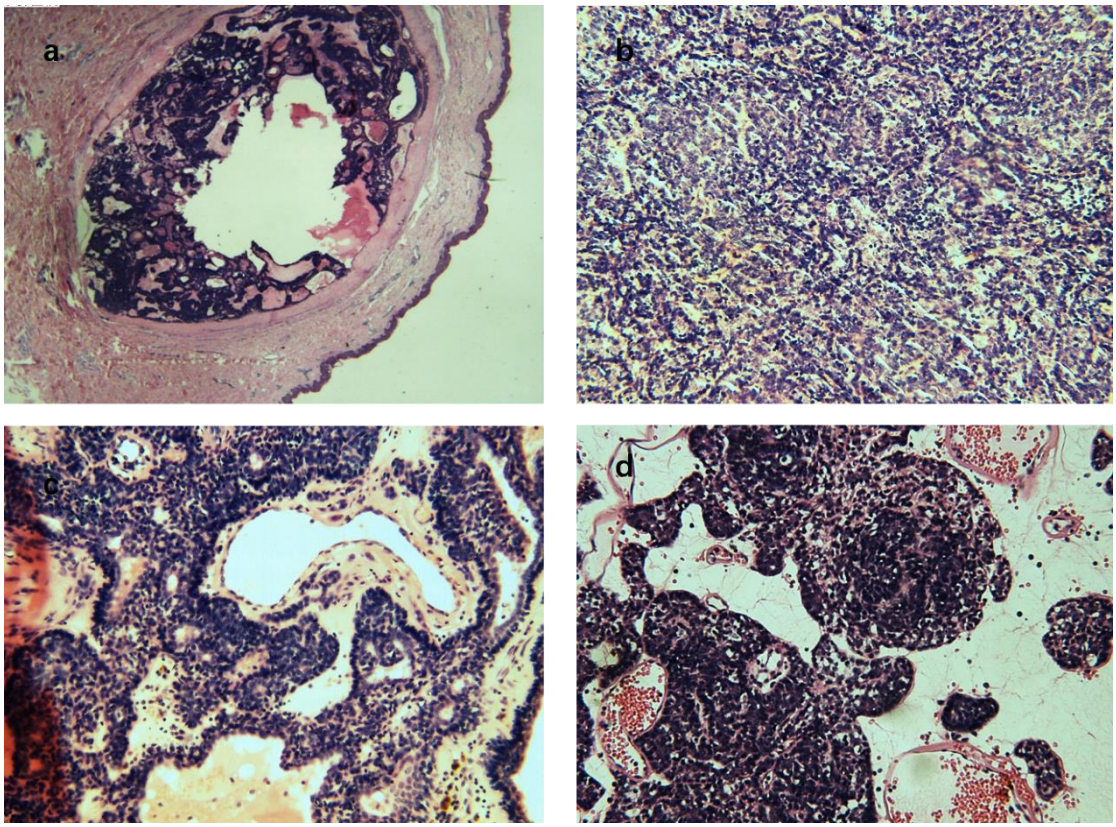
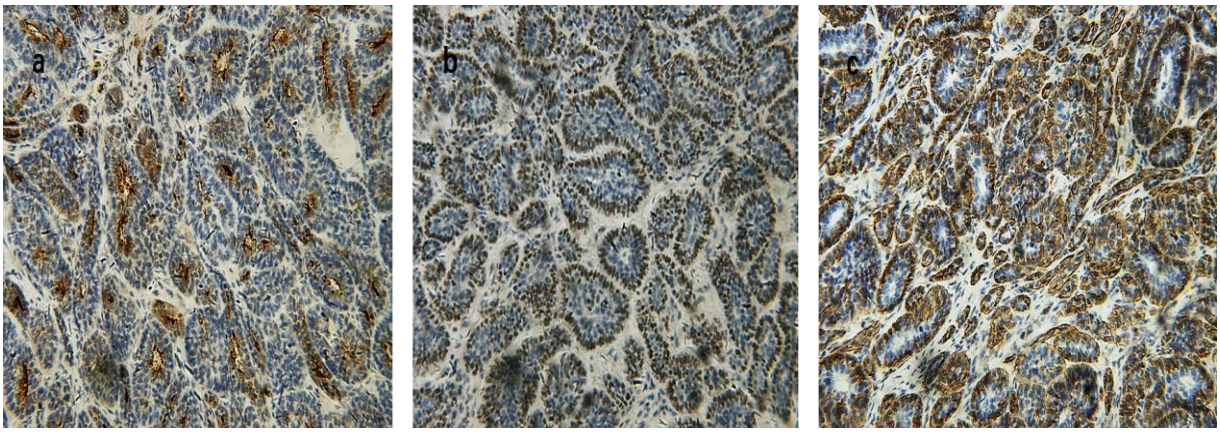


Figure 2. *(a-d): Hematoxylin & Eosin-stained: a: the tumor is located within the dermis and surrounded by the capsule (HE × 40); b: tumor cells showing rosette-like arrangement of the two-cell population: small dark and basaloid cells with hyperchromatic nuclei and larger cells with a pale nucleus (HE × 100); c: prominent deposition of eosinophilic basement membrane-like material and focal duct formation were noted (HE × 200); d: vascular lumen filled with red blood cells (HE × 200).*



a b c

Figure 3. *(a-c): Immunohistochemical -stained: positive staining: a: EMA: expression of sweat gland lumen marker highlighted the central cells (IHC × 200); b-c: SMA, p63: expression of myoepithelial marker highlighted the small-sized cells with dark chromatin (IHC × 200).*

4. Discussion

SA is more common in middle-aged and young patients [6], with no gender difference. SA commonly occurs at head and neck, followed by the trunk and limbs [7]. It can also be seen in rare areas such as the perianal [6], nipple, and auricle [8, 9]. These tumors usually present as a single, tender, well circumscribed intradermal nodule [7]. When we observed multiple lesions, the occurrence of BSS should be considered [10]. In this study, tumors were more common in the trunk. Except for one case with multiple nodules, the rest were single nodules, similar to literature reports, with an average age of 41 years, older than foreign reports. According to the re-

ports that SA can also be multiple, appearing linear [11] or arranged along the blaschko line [12].

In clinical, SA often presents as a single nodule, partially accompanied by pain, so it is easily confused with fibroids, glomus tumors, etc. Some cases may apear blue or red, they are easily misdiagnosed as blue nevi, vascular sarcoma, melanoma, etc. In this study, 11 cases were clinically diagnosed with one case of lipoma, 3 cases of epidermal cyst, and 7 cases of skin tumor (not specifically classified). Therefore, SA relies on pathological features to confirm the diagnosis. The histological characteristics of SA: The epidermis is generally normal, and the tumor is located within the dermis, separated by a fibrous capsule of varying thickness, with clear boundaries. Tumor cells are densely distributed, mainly composed of two types of cells: Surrounded by small cells with dark nucleus and larger cells with pale nucleus in the center. Intertwining cords around connective tissue and trabecules can be detected and a large number of lymphocytes gathering in the stroma. Differential diagnosis: (1) Glomangioma: When there is obvious dilation of blood vessels, it should be differentiated from glomangioma, but it has no ductal, the lumen is composed of a single layer of endothelial cells, and the tumor cells are composed of uniform circular cells. (2) Basal cell carcinoma: tumor cell masses are located in the dermis, small and dark nucleus, but the surrounding cells are arranged in a palisade like pattern with obvious mitotic figures.

Although the incidence of malignant transformation from SA to spiradenocarcinoma is low, high-grade carcinomas show the high recurrence and mortality rates [13]. When there is a tumor that grows rapidly in the short term, accompanied by obvious tenderness, ulcers, and bleeding, we should be alert to the possibility of malignancy [14]. The key to diagnose spiradenocarcinoma is the presence of SA components [15]. Under the microscope, spiradenocarcinoma is mainly manifested in two forms: one is the gradual transformation from benign to malignant, with visible transitional zones between benign and malignant, and two types of tumor cells transforming into similar uniform atypical cells. Another type present there is no obvious transition zone, which can differentiate into sarcomatoid carcinoma, squamous cell carcinoma, adenomatous ductal carcinoma, etc [16]. Suspicion of spiradenocarcinoma should lead to performance of a magnetic resonance imaging and fluorodeoxyglucose-positron emission tomography to establish tumor extent [17]. According to the latest report, PD-1 Inhibition can be used to treat metastatic spiradenocarcinoma [18].

The diagnosis of SA mainly relies on histopathology, and immunohistochemistry can serve as an auxiliary diagnosis. EMA and CEA mark the sweat gland lumen structure, positive expression of SMA and P63 indicates their differentiation into myoepithelial cells.

5. Conclusion

In summary, the clinical manifestations of SA are not specific and are prone to misdiagnosis or missed diagnosis, relying on clinicopathologic to confirm. The biological behavior of SA is often benign, and complete surgical resection is the preferred treatment. When the course of the disease is long or relapses, we should be alert to the possibility of malignancy.

Abbreviations

SA: Spiradenoma
BSS: Brooke-Spiegler Syndrome
HE: Hematoxylin & Eosin-Stained
IHC: Immunohistochemical-Stained
EMA: Epithelial Membrane Antigen
SMA: Smooth MUSCLE Actin

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Author Contributions

Qianyuan Tian: Conceptualization, Data curation, Resources, Investigation, Writing - original draft, Writing - review & editing
Yunfeng Hu: Supervision, Writing - review & editing
Yuanzhi Lu: Supervision, Writing - review & editing

Conflict of Interests

The authors declare no conflict of interest.

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