Adnexal Tumor of The Arm: A Rare Case Report and Review of Literature

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Dear Editor,

Enclosed is a manuscript, titled “Adnexal tumor of the arm: A rare case report and review of literature” for your consideration. This localization is rare, and this article is the first report from our institution. I confirm that it has not been published and it is not under consideration elsewhere. I confirm also that human subjects in this study were treated in accordance with standard ethical guidelines. Thank you for your time and consideration; I look forward to hearing from you.

Competing Interests:
The authors declare that they have no competing interests.
Adnexal Tumor of The Arm: A Rare Case Report and Review of Literature

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Abstract

Introduction:
Adnexal tumors are rare neoplasms and account for 1-2% of soft tissue tumors. They can be of pilair, sebaceous or sweat component. Several locations have been described (eye, eyelid, nose ...), location in the limbs is exceptional.

Case presentation:
We report the case of a 72 years old Moroccan woman, presenting since a year a nodule at the left inner arm. A complete excision of the tumor was performed. The evolution is marked by recurrence of the scar in the form of two mobile nodes with ipsilateral axillary lymphadenopathy. The patient benefited of a wide excision taking the nodules and the old scar associated with a homolateral axillary dissection. The analysis of the surgical specimen revealed a rare tumor type Eccrine Spiradenoma. After five years, the patient is free from local recurrence or metastases.

Conclusion:
The Eccrine Spiradenoma is benign tumor but often do not overlook the risk of malignancy. Its management must be early, radical surgery is the treatment of choice, the place of radiotherapy and chemotherapy in the treatment of malignant tumors is controversial.

Introduction

Adnexal tumors (AT) are benign tumors with good prognosis. They are numerous: Their classification is complex, only a precise histological examination can differentiate each type [1]. The AT can be classified into tumors that can be isolated of sweat, sebaceous or pilair component. They can be multiple tumors often autosomal dominant, or tumors associated to other cancers associated with visceral or multiple syndromes such as BROOKE SPIEGLER syndrome [1]. We report a particular original AT originates from sudoral component named Eccrine Spiradenoma (ES). The ES is a usually benign tumor that may recur locally in case of incomplete excision and whose appearance is specific.

Discussion

First described by Sutton in 1934 [2] and by Kersting and Helwig in 1956 [3-4], the ES is a benign tumor dysembryoplastic developed in the secretory portion of eccrine sweat deep glands. The ES can be seen at any age [2], but it is especially common in young adults without sex predilection [3]. Clinically the ES occurs most commonly as a solitary firm nodule (79%), flesh-colored or bluish red, located in the dermis or hypodermis. Its diameter is variable (0.5 cm to 5 cm) [2-3]. The most common sites are the face, neck, trunk, rarely in the extremities. These nodules are either spontaneously or painful on palpation (23%), pain is mainly due to small demyelinated axons that permeate the mantle hyalinized stroma [2],
whereas, in carcinoma, pain is due to neural invasion by the tumor cells. Multiple forms can be seen. They may be isolated (sporadic) or associated with family trichoepitheliomias and cylindromas through Rasmussen's syndrome [3]. Degeneration of Spiradenoma to a carcinoma is reported by several authors [5], emphasizing the fact that degeneration is an exceptional phenomenon [6-7]. Malignant degeneration should be suspected in a recent morphological change such a rapid increase in tumor size, a color change or ulceration of the underlying epidermis. The definitive diagnosis is histological. Skin biopsy confirmed the diagnosis of ES, macroscopically it appears as a lobulated dermal tumor formation bounded by a fibrous capsule with no connection to the epidermis. Histologically the tumor is characterized by basophilic lobules made ??of two juxtaposed cell component: small cells in the periphery of the lobules or palisade around vessels and large clear cells in the center of the nodules sometimes forming tubular structures [6]. They may sometimes contain an eosinophilic секретory material Alcian Blue and PAS positive/diastase resistant [6-8]. The stroma is often focally hyalinized cylindromatous aspect. When the vessels are prominent, it carries the variant of vascular ES. The cystic variant is a rare one; it results from ductal ectasia secondary to intra tumoral obstruction. Other aspects more or less characteristic are described, such as presence of lymphocytes cells around vessels or peritumoral oedema [6]. Immuno histochemistry is a great help for both the positive and differential diagnosis such as to the pathogenesis [6]. Small cells and clear cells specifically express keratins [6]. In addition to clear cells lining the tubular structures show intense apical membrane staining with anti-EMA (epithelial membrane antigen) and anti-CEA (carcinoeembryonic antigen), not present in other AT [9]. Another tumor marker known as GCDFP-15 (Gross Cystic Disease Fluid Protein-15), representing a protein originally identified in cystic breast disease, is also found in the ES [10]. S100 protein is expressed by a subpopulation of cells eccrine sweat [10]. Epithelial cells also express the ES Leu 7 antigen, ferritin, lysozyme [16] and the oncoprotein bcl-2 [11]. Table 1 shows the expression of these markers in the variants of AT. The differential diagnosis is mainly cylindromas, histologically very similar tumors, sometimes associated, which does include architectural differences. Also basal cell carcinoma can be a differential diagnosis. The ES merges with richly vascularized tumors as benign glomangioma. However, all these tumors are devoid of cell maturation characteristic of the ES [6]. Other tumors or entities may be associated with the ES, it is most often a trichoepithelioma or hidradenoma and more rarely an epidermal cystic or dermato fibroma [12]. The origin and differentiation of ES are still controversial [6]. For Watanabe et al. the ES would derive from the transition zone between the pack and secretory excretory duct coiled sweat gland [13]. Surgery remains the mainstay of treatment [6], it consists of a wide resection. The margins of resection must be considered: lateral margins must be at least 3 cm and 1 cm in depth[14]. Dissection is made necessary when lymph node is observed [14]. CO2 laser or radiotherapy may be considered as interesting alternative in multiple and extensive forms [12-13]. The ES is a benign tumor and deemed to have a good prognosis when surgical resection is radical. Recurrence is exceptional; it is frequent when the resection is marginal. The recurrence was treated by wide re-excision [2]. In the case of malignant transformation of ES, surgical treatment must be aggressive [2], with a poorer prognosis because of the high risk of recurrence and metastasis [15].

Conclusion

The ES is a rare tumor; the arm localization is even rarer. This is a benign tumor of slow evolution that involves a risk of malignant degeneration especially if treatment is not radical. The diagnosis is essentially histological; the treatment of choice is radical surgery. CO2 laser or radiotherapy may be offered in extensive and disseminated forms. More diagnosed is early and more surgery is extensive more prognosis is better. Regular monitoring is necessary to detect local recurrence, locoregional, or metastatic spread.

References

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Illustrations

Illustration 1

Histological aspect of Eccrine Spiradenoma: aspect of basaloid islands and bays lined by two layers of cells with basophilic and regular nuclei.
Illustration 2

Table: Immunohistochemical phenotype of adnexal tumors (CEA: carcinoembryonic antigen, EMA: epithelial membrane antigen, GCDFP-15: gross cystic disease fluid protein)

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