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SYRINGOCYSTADENOMA PAPILLIFERUM - A RARE CASE REPORT

Pathology

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ABSTRACT

Syringocystadenoma papilliferum is a rare benign adnexal tumor of the apocrine or the eccrinedifferentiation. It is usually appears at birth or during puberty occurring most commonly on scalp and face. The clinical presentation varies from nodular swelling to ulcerated lesions.Cytological diagnosis of this entity is rare. We present a rare case syringocystadenoma papilliferum at a rare site diagnosed on cytology and confirmed on histopathological examination.

KEYWORDS

Adnexal tumor, syringocystadenoma papilliferum, SCAP, fine needle aspiration cytology

INTRODUCTION

Syringocystadenoma papilliferum (SCAP) is a rare adnexal tumor of unknown etiology [1]. It is classified as a benign tumor of apocrine or eccrinedifferentiation. [2] It is first noted at birth or in early childhood and presents as a papule or several papules in a linear arrangement or as a solid plaque. It frequently arises around puberty within a nevus sebaceous that has been present since birth. It most commonly occurson face and scalp. The cytological diagnosis of this entity is uncommonly reported. We present a case of SCAP in unusual location in 34 year old male diagnosed on cytology and confirmed on histopathological examination.

Case Report

A 34-year old male presented with painless, slowly growing nodular swelling on his left axilla for one year. Physical examination revealed well circumscribed, mobile, non-tender nodular swelling approximately measuring 2.5x2cm. The overlying skin was unremarkable. Fine-needle aspiration cytology of the lesion was done using 23 gauge needle and 10ml disposable syringe.Smears were prepared and stained with Giemsa stain.

The smears were moderately cellular comprising of cohesive clusters of epithelial cells along with singly dispersed cells.Occasional papillary formations are also seen. The individual cells were plasmacytoid cells having moderate amount of dense basophilic cytoplasm with eccentric round nuclei with mild nuclear atypia. The background was clear. [Figure 1]

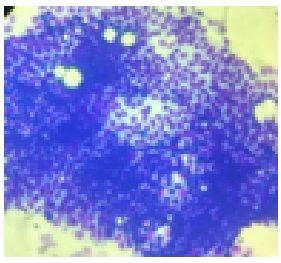

Figure 1: Photomicrograph showing cohesive clusters of benign epithelial cells in FNAC smear (Giemsa, X40).

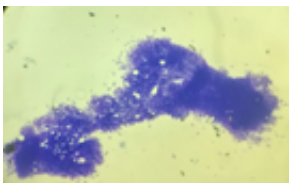

Figure 2: Photomicrograph showing papillary formation by benign epithelial cells in FNAC smear (Giemsa, X10).

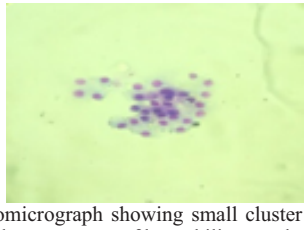

Figure 3: Photomicrograph showing small cluster of plasmacytoid cells having moderate amount of basophilic cytoplasm with eccentric round nuclei. The background is clear. (Giemsa, X40).

Surgical excision was done and specimen was sent to histopathological examinationGross examination revealed single, partly skin covered, grey white soft tissue mass measuring 3x3x0.5 cm. Cut surface showed yellow and grey white areas (Figure 4).

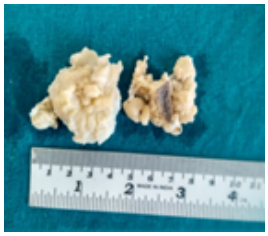

Figure 4: Gross appearance showing partly skin covered, grey white soft tissue mass.

Microscopic examination showed epidermis showing varying degrees of papillomatosis. Multiple cystic invaginations were seen extending downward from the epidermis. The invaginations were lined by squamous, keratinizing cells. The papillary projections and the lower portion of the invaginations were lined by glandular epithelium consisted of two rows of cells. The luminal row of cells consisted of high columnar cells with oval nuclei and faintly eosinophilic cytoplasm. Some of these cells showed active decapitation secretion and cellular debris in the lumina. The outer row of cells consisted of small cuboidal cells with round nuclei and scanty cytoplasm. The connective tissue core was filled with plasma cells, lymphocytes and dilated capillaries. The histopathological findings were consistent with the diagnosis of syringocystadenoma papilleferum (Figure 5). The post-operative period was uneventful. The patient was on regular follow-up and was doing well, thereafter.

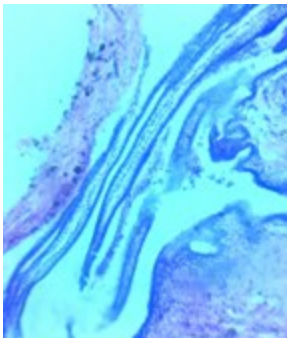

Figure 5- Photomicrograph showing papillary projections lined by double epithelial layers and lumen filled with dense plasma cell infiltrate (Inset) (H& E, X40).

DISCUSSION

Stokes in 1917 first described the SCAP under the term *nevus syringadenomatosuspapilliferus*, since then SCAP has been increasingly

reported in the English literature[3]. Syringocystadenoma papilliferum presents as brownish or erythematous papules or nodules, hairless plaques of varying sizes which range from 5 to 160 mm. Their surfaces can be smooth, flat, papillomatous, or verrucous. Increase in their sizes, crustations, nodular or verrucous transformations is noted at puberty. Most of the patients present with solitary lesions. Multiple lesions are usually associated with nevus sebaceous. Both sexes are equally affected [4].In about 75% of cases, SCAP arises within a preexistent organoid sebaceous nevus of Jadassohn.The remaining 25% of SCAP arise on the trunk and genitocrural region during adolescence or adult life without a preexisting lesion, usually in the solitary nodular form. In a large series of 100 cases reported by Helwig and Hackney 55 were on the scalp, 11 on the forehead and temple, 5 on the face and 3 on the upper lip [5].

Transition to carcinoma is rare and malignant change is heralded by a rapid increase in size, appearance of new lesions, bleeding and appearance of metastatic lymph nodes. In about one-tenth of cases of SCAP, basal cell carcinoma can secondarily develop [5]. Uncommon sites of occurrence which have been reported in the literature include chest, arms, breast, eyelids, axilla, scrotum, thigh, lower legs, toes, inguinal and perineal regions [6].Ninety percent of the cases show involvement of anatomic sites which are normally devoid of apocrine elements [7].Nodular variety shows more involvement of the trunk.[4].The differential diagnosis of SCAP on histology includes hidradenoma papilliferum, papillary eccrine adenomas, warty dyskeratoma, tubular apocrine adenoma and inverted follicula keratosis.In our case absence of acantholytic, dyskeratotic cells and keratinous material exclude the diagnosis of inverted follicular keratosis. Hidradenoma papilliferum and papillary ecrine adenoma usually yield a mucoid aspirate and show the basaloid cell in the background of protienaceous fluid and are thus excluded. Tubular apocrine adenoma on cytology shows predominantly cuboidal and columnar cells with or without secretions. In our case presence of plasmacytoid cells in a clear background clinches the diagnosis.

The treatment for Syringocystadenoma papilliferum is excision biopsy, which also confirms the diagnosis. CO_2 laser excision of SCAP of the head and neck is a treatment option in anatomic areas that are unfavorable for excision and grafting[8]. SCAP has also been successfully treated with Mohs micrographic surgery[9].

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