Case Report

Cytological diagnosis of syringocystadenoma papilliferum a rare adnexal tumor

ABSTRACT
Syringocystadenoma papilliferum is an uncommon benign adnexal tumor occurring most commonly on scalp and face. The clinical presentation varies from nodular swelling to ulcerated lesions. The lesion was on a rare site (eyebrow) was diagnosed cytologically.

Key words: Adnexal tumor; fine needle aspiration cytology; syringocystadenoma papilliferum

Introduction
Syringocystadenoma papilliferum (SCAP) is a hamartomatous adnexal tumor with unknown etiology. It is classified as a benign tumor of apocrine differentiation. It either develops independently or is commonly seen in association with the nevus sebaceous. Almost half are present at birth or appear during infancy. It commonly arises around the time of puberty or less commonly during adolescence or adult life. Cytological diagnosis of this entity is uncommonly reported. We reached a cytological diagnosis of this rare entity at a rare site (eyebrow) and hence this report.

Case Report
A 35-year-old female patient presented with a lesion on the medial end of left eyebrow. She had a small nodular elevated swelling for the last 2 months. There was no history of itching, fever, weight loss, or other constitutional symptoms.

Physical examination revealed a solitary, well circumscribed swelling of 1 cm diameter. The swelling was firm in consistency, mobile and nontender. The skin over swelling was normal. Rest of the cutaneous and general physical examination was unremarkable. History was insignificant.

Fine-needle aspiration cytology of the lesion was done using a 23G needle, and a provisional diagnosis of SCAP was made. The lesion was then excised and submitted for biopsy.

Fine-needle aspiration cytology smears were highly cellular comprising of epithelial cells forming cohesive aggregates as well as dispersed as single cells. However, no true papillary fragments were seen. The cells had a moderate amount of dense cytoplasm with eccentric round nuclei and mild nuclear atypia. Picture was suggestive of benign adnexal tumor-SCAP [Figure 1].

The tumor was excised, and a small grey white soft tissue piece measuring 0.8 cm in size was received for histopathological examination. Sections showed presence of numerous ducts lined by flattened cells and surrounded by round to polyhedral plasmacytoid cells arranged in nests and cords. Papillae formation was seen at places and showed infiltration by few plasma cells consistent with the diagnosis of SCAP [Figure 2].

Discussion
Since the first description by Stokes in 1917 under the term nevus syringadenomatosus papilliferus, SCAP has been increasingly reported in the English literature.
Syringocystadenoma papilliferum is an uncommon benign adnexal tumor occurring most commonly on scalp and face. In about 75% of cases, SCAP arises within a preexistent organoid sebaceous nevus of Jadassohn.\[5\] 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. Lesion in the present case arose in the same manner.

In a large series of 100 cases reported by Helwig and Hackney,\[5\] 55 were on the scalp, 11 on the forehead and temple, 5 on the face and 3 on the upper lip, but none was reported on the eyebrow.

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.\[9\]

The differential diagnosis of SCAP includes hidradenoma papilliferum, papillary eccrine adenomas, warty dyskeratoma, tubular apocrine adenoma and inverted follicular keratosis.

Absence of acantholytic, dyskeratotic cells and keratinous material exclude the diagnosis of inverted follicular keratosis. Hidradenoma papilliferum and papillary eccrine adenoma usually yield a mucoid aspirate and show the basaloid cell in the background of proteinaceous 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 only treatment for SCAP is excision biopsy. 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.\[6\] SCAP has also been successfully treated with Mohs micrographic surgery.\[7\]

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