Single Case

Syringocystadenoma Papilliferum: A Case Report and Review of the Literature

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Keywords

Syringocystadenoma papilliferum · Apocrine adenoma

Abstract

Syringocystadenoma papilliferum is a very rare, benign adnexal tumor that originates from the apocrine sweat glands. Herein, we report a 25-year-old male who presented with a 10-year history of an asymptomatic slowly growing skin lesion on his face. Skin examination revealed a solitary rounded 3 × 3 cm erythematous plaque with central crustation on the right side of his face. Punch skin biopsy was taken from the lesion. The epidermis showed downward papillomatous extensions. The dermis showed multiple epithelial sheets and dilated ducts that were lined by columnar cells. On the basis of the above clinicopathological findings, the diagnosis of syringocystadenoma papilliferum was made. The patient was reassured and referred to a surgeon for surgical excision of the lesion.

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Introduction

Syringocystadenoma papilliferum (SP) is a very rare, benign adnexal tumor that originates from the apocrine or eccrine sweat glands. However, in rare cases, SP may convert into a malignant lesion [1]. SP is common during childhood or early adulthood and occurs as asymptomatic slowly growing crusted reddish papule or plaque. SP is rarely pigmented. The

most common location is the scalp and face. It rarely occurs on the nipple, breast, genitalia, eyelids, and extremities [1–3]. SP can be single or multiple. The cause of SP is not clear. One-third of SP arises in nevus sebaceous [1, 2]. BRAF V600E mutation or activating mutations in HRAS (or less often KRAS) have been reported in sporadic forms, whereas nevus sebaceous carries the same HRAS mutation as the underlying nevus [1].

Case Report

A 25-year-old male presented with a 10-year history of an asymptomatic slowly growing solitary skin lesion on his face. The lesion sometimes oozed fluid or became ulcerated or bled. He had been using several topical and systemic steroids, antibiotics, and antifungals for years but without improvement. Past medical history, family history, and review systems were unremarkable. Skin examination revealed a solitary rounded 3 × 3 cm fleshy plaque with central crusting on the right side of his face (Fig. 1). There was no lymphadenopathy. Punch skin biopsy was taken from the lesion. The epidermis showed downward papillomatous extensions. The dermis showed multiple epithelial sheets and dilated ducts that were lined by columnar cells (Fig. 2). On the basis of the above clinicopathological findings, the diagnosis of SP was made. The patient was reassured and referred to a surgeon for surgical excision of the lesion.

Discussion

SP is a rare benign adnexal neoplasm that originates from either apocrine or eccrine sweat glands. The main differential diagnosis in our case includes basal cell carcinoma, cutaneous lymphoma, factitious dermatitis, and pyogenic granuloma. However, slowly growing fleshy plaque that has been growing for years and sometimes oozes fluid and sometimes bleeds is characteristic for SP. Syringocystadenocarcinoma papilliferum, which is the malignant counterpart of SP, should be ruled out when evaluating the histopathology of SP [4]. Basal cell carcinoma has been reported to be seen in 10% of patients with SP. So, a patient with SP should be evaluated for associated basal cell carcinoma. The treatment of choice of SP is surgical excision. However, recurrence is common [1, 2, 5]. Our case was treated by surgical excision, with no recurrence until the time of the report.

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Statement of Ethics

The authors have no ethical conflicts to disclose. Written consent has been obtained from the patient.

Disclosure Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Conception and design of the study: Khalid Al Hawsawi. Data collection: Amani Alharazi. Introduction and discussion: Amani Alharazi, Abeer Ashary, Asmaa Siddique. Drafting the manuscript: Amani Alharazi, Abeer Ashary, Asmaa Siddique. Revising the manuscript critically for important intellectual content: Khalid Al Hawsawi. Approval of the version of the manuscript to be published: Khalid Al Hawsawi.

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Fig. 1. Solitary rounded 3 × 3 cm erythematous plaque with central crusting on the right side of his face.

Fig. 2. Punch skin biopsy from the lesion. The epidermis shows downward papillomatous extensions. The dermis shows multiple epithelial sheets and dilated ducts that are lined by columnar cells.