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Case Report

Syringocystadenoma Papilliferum of the Right Thigh in An Adolescent Girl

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Abstract

Syringocystadenoma papilliferum is an uncommon, benign adnexal tumor of eccrine or apocrine origin, often appearing at birth or before puberty. Lesions may occur spontaneously or emerge from a pre-existing sebaceous nevus. We report the case of a 15-year-old girl who arrived with an indurated, multilobulated cystic tumour on the right thigh. Excisional biopsy revealed the hallmark histological characteristics of syringocystadenoma papilliferum. The interest of this case report rests in the rarity of syringocystadenoma papilliferum and its atypical presentation on the right thigh.

Introduction

Syringocystadenoma Papilliferum (SCAP) is an uncommon, benign adnexal tumor of uncertain origin that is thought to originate in the apocrine or eccrine glands. Although occurrences in other anatomical locations are less frequently reported, it is most commonly encountered in pediatric and adolescent populations, with symptoms typically presenting on the scalp, face, and neck. Histopathological examination is required to identify SCAP from other dermatoses and establish the best course of treatment [1,2].

Case Presentation

We discuss the case of a 15-year-old female with no known medical history who arrived with a big tumor on the anteromedial portion of her right thigh. This pathology has existed since early childhood, and the patient sought medical attention due to an increase in size.A clinical examination revealed a pedunculated, vegetative, erythematous tumor measuring 18x18 mm, with a keratotic surface, uneven boundaries, and nodules evident at the periphery **(Figure 1)**.

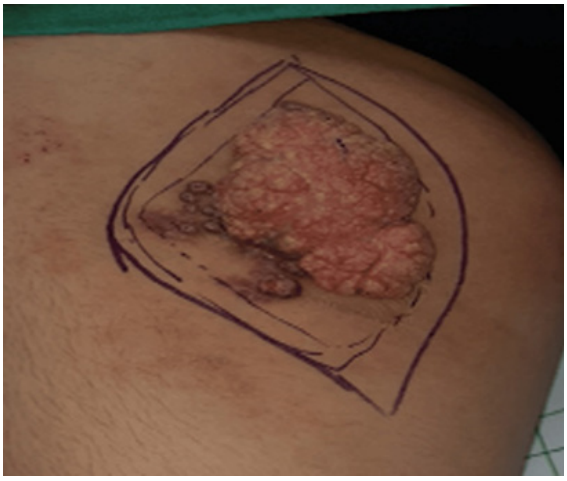

Figure 1: An image showing the erythematous mass on the anteromedial aspect of the thigh.

It was decided to proceed with resection for pathological diagnosis due to the mass's size.

The patient received an immediate excision of the lesion with 1cm broad margins **(Figure 2)**.

The advancement was characterized by suture dehiscence after tension closure, promting the implementation of a guided tissue healing strategy **(Figure 3)**.

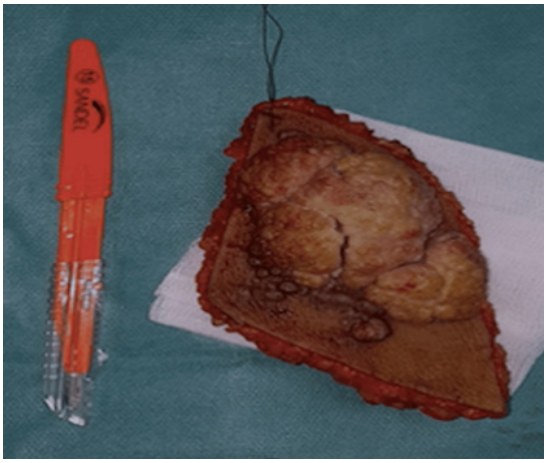

Figure 2: Gross image showing excised mass measured at 18 x 18 mm.

Histological examination of the mass showed partially cystic structures lined by several layers of columnar cells with eosinophilic cytoplasm at the apical pole with decapitation secretion. The stroma had a local mononuclear leukocytic infiltrate

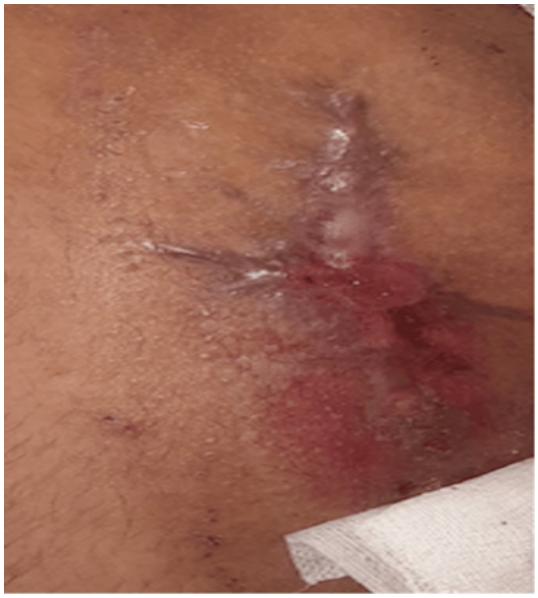

Figure 3: Six month progression of a healing scar.

rich in plasma cells with no suspicious lesions **(Figure 3)**. A final diagnosis of SCAP was entertained. The evolution was favorable with no recurrence after two years of follow-up **(Figure 4)**.

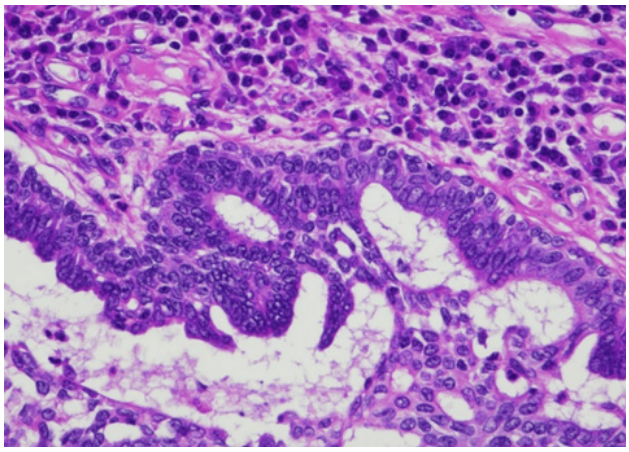

Figure 4: Microscopic features of Syringocystadenoma papilliferum.

Discussion

The specifics of our case are found in the rarity of the SCAP and the extent of localization at the thigh.

Beginning from pluripotent cells [1,2], SCAP is a hamartomatous adnexal tumor. Derived from apocrine and eccrine glands, its histogenesis is still debated [3]. It is accepted that differentiation is predominantly apocrine [1]. It is a childhood or adolescence neoplasm, observed since birth in 50% of occurrences, as in the present case.

Its clinical presentation is a papule, plaque or a solitary or aggregated nodule, with no hairs, asymptomatic, but that may become exsudative and with a linear aspect arrangement [1,4]. It has a tendency to develop in size during adolescence, becoming more verrucous and papillomatous [1,3]. Its most prominent places are the scalp, neck and face. In the investigation carried out by Mammino and Al 75% were located at the level of the head and the neck 20% were located next to the neck and 5% at the level of the extremities [5].

SCAP on the thigh is uncommon. Eight examples were identified by a thorough study of the literature [5], including one tumour that developed in geant comedo and several linear lesions [6]. Recently, a thigh lesion without a link on the underlying epidermis was discovered by Yamamoto and Mamada. The middermia contained the nodule, and the epidermis above it was unbroken and free of acantosis [7]. Remarkably, Stokes discovered the first instance of SCAP on the thigh in 1917.

It is frequently associated with hamartomas originating from sebaceous or follicular glands. Syringocystadenoma papilliferum is linked to a sebaceous nevus in approximately one-third of cases. A syringocystadenoma papilliferum may be one of several adnexal tumours that have been observed to develop on a sebaceous nevus, including trichoblastoma apocrine adenoma, hidradenoma pappilliferum, poroma folliculare, trichilemmoma, and others [1,2,4].

According to histology, it presents as a dermal endophytic tumour with irregular scaly epithelial papillary projections that create ductile structures that link to the surface. These ductile structures are aligned by glandular epithelium, which is made up of an inside layer of cylindrical cells with plasmocyte-rich inflammatory infiltration and decapitation secretion, and an exterior layer of cuboid cells with round nuclei and scant cytoplasm [1,2].

Tumor cells demonstrate a reaction of positive staining with carcinoembryonic antigen. It is rarely associated with malignant progression, however basal cell carcinoma development was observed in 10% of the cases, notably when paired with sebaceous nevus of Jadassohn. Squamous cell carcinoma and syringocystadenocarcinoma papilliferum were also recorded as a progression of SCAP, but are extremely rare. Healing The treatment for Syringocystadenoma papilliferum is excision biopsy , which also supports the diagnosis [8,9], but there are examples in which removal was effective using CO2 laser in places undesirable for surgery [10].

Conclusion

The interest of this case report rests in the rarity of syringocystadenoma papilliferum and its atypical presentation in the thigh. Although benign, it is vital to distinguish syringocystadenoma papilliferum from other skin tumors to enable an accurate diagnosis and treatment. The overall risk of contiguous malignant squamous proliferations in syringocystadenoma papilliferum is rare.

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