Syringocystadenoma papilliferum combined with a tubular apocrine adenoma*

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Abstract: Syringocystadenoma papilliferum and tubular apocrine adenoma are rare benign sweat gland tumors. Syringocystadenoma papilliferum occurs alone or in association with other tumors. Although it is rare, the association of tubular apocrine adenoma with syringocystadenoma papilliferum developing in a sebaceous nevus on the scalp is well documented. However, the co-existence of these two tumors without the background of a sebaceous nevus has not been frequently reported. Syringocystadenoma papilliferum and tubular apocrine adenoma may have a histopathological overlap, but a few cases of a syringocystadenoma papilliferum combined with a tubular apocrine adenoma have been reported. Herein we describe an unusual case of syringocystadenoma papilliferum co-existing with a tubular apocrine adenoma located on the back of a 14-year-old patient in the absence of a pre-existing sebaceous nevus.
Keywords: Adenoma, sweat gland; Nevus sebaceous of Jadassohn

INTRODUCTION

Syringocystadenoma papilliferum (SCAP) and tubular apocrine adenoma (TAA) are rare and benign sweat gland tumors.[1]

SCAP may occur alone or in association with an organoid nevus (sebaceous nevus). In both instances, it is most often found on the scalp or face, occurring in other locations in only a quarter of cases. It is rarely found on the trunk or limbs and occasionally coexists with other tumors.[2]

In some cases, TAA may develop in association with SCAP, which occurs in the superficial portion of the TAA. However, the combination of these two tumors in the absence of a sebaceous nevus and affecting another area other than the scalp has not been frequently described.[2]

We report a case of SCAP associated with TAA on the back, in the absence of a preexisting sebaceous nevus, affecting an adolescent patient. The objective of the present report is to emphasize the unusual location and the rare association of these two tumors in the absence of sebaceous nevi. We also emphasize the importance of the differential diagnosis, as well as the adequate treatment of this rare disease.

CASE REPORT

A 14-year-old white male patient, asymtomatic congenital lesion with recent increase in size and sporadic discharge.

Physical examination revealed a whitish nodule with central exulceration with a yellowish exsudate, sitting on a slightly infiltrated base in the interscapular area. The lesion was completely excised (Figure 1).

On histopathological examination, the superficial component was characterized by cystic invaginations of the infundibular epithelium projecting into the dermis, covered by a double cell layer: the innermost layer revealed columnar cells, with the typical decapitation secretion; and the outermost layer, cuboidal cells with papillary projections. The upper dermis also revealed plasma-cell rich infiltrates (Figures 2 and 3).

In the deep portion, we observed glandular structures of varied sizes, also presenting a double layer of epithelial cells. The findings on the superficial portion of the lesion were interpreted as SCAP, and those found in the deep layers, as TAA (Figure 4).

We observed no recurrence after 4 months of follow-up.

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DISCUSSION

Syringocystadenoma papilliferum (SCAP) is a benign sweat gland tumor noted at birth or early childhood that presents itself as a solitary papule, papules in a linear distribution, or plaques. The lesion increases in size at puberty, becoming papillomatous and crusted. It occurs most commonly on the scalp and face, but in 25% of cases, it can occur in any area of the body (trunk, vulva, and limbs). Its apocrine differentiation can be demonstrated by the presence of columnar epithelium with decapitation secretion.[1,2]

Tubular apocrine adenoma (TAA) is also an unusual sweat gland tumor. It is usually found in adults as a solitary nodule located most often on the scalp.[1]

The association of SCAP and TAA was first described by Toribio et al.[4] in 1987. Since then, a few similar cases have been described in the literature, with rare cases described in the absence of a sebaceous nevus and affecting other areas of the body. We found only 12 cases of SCAP associated with TAA in the literature, and only three cases describe the absence of preexisting sebaceous nevi (Table 1).[1,3]

Histopathologically, SCAP consists of cystic invaginations that project into the dermis and open onto the skin surface through one or more orifices. It consists of a double layer of cells: an inner

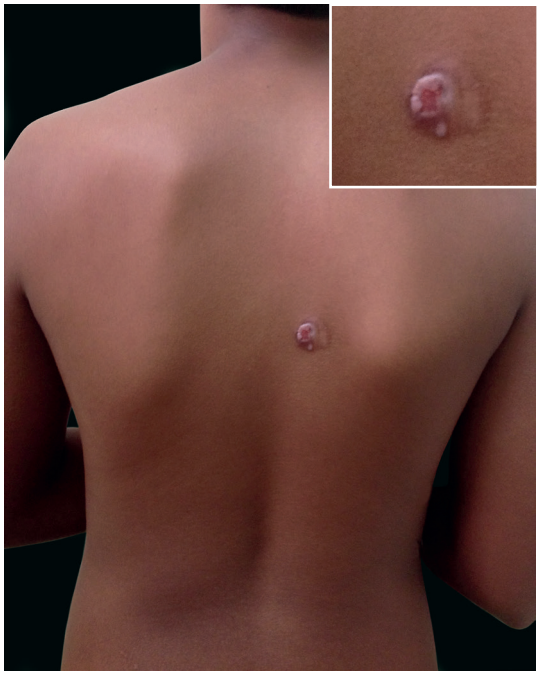

Figure 1: A whitish nodule with central exulceration, sitting on a slightly infiltrated base in the interscapular area

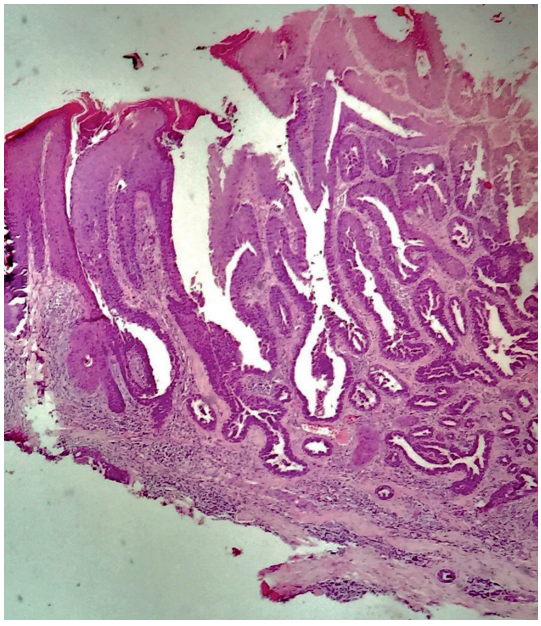

Figure 2: Upper part of the tumor showing cystic invagination extending downward from the epidermis (SCAP) and lower portion showing islets of tubular structures of varying sizes in the deep dermis (TAA) (Hematoxylin & eosin, X4)

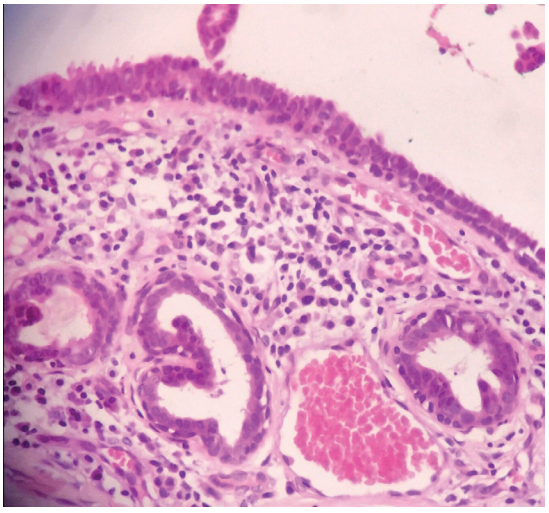

Figure 3: SCAP: double layer of cells. The inner layer is composed of columnar cells, where decapitation secretion is observed, and the outer layer is formed by cuboidal cells. Stroma with infiltrate rich in inflammatory cells, mainly plasma cells. (Hematoxylin & eosin X40)

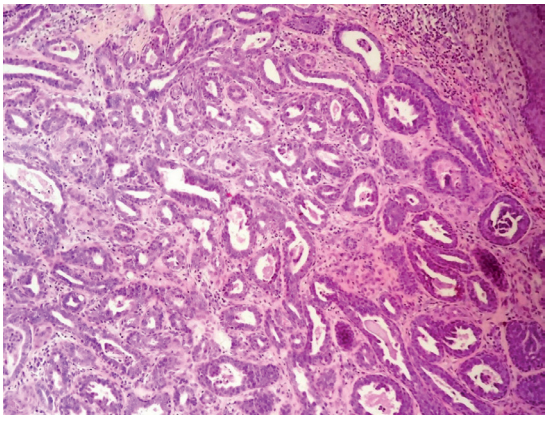

Figure 4: TAA exhibiting numerous irregular-shaped tubular structures with a dual cell layer. The tubules are embedded in a fibrous paucicellular stroma (Hematoxylin & eosin X10)

Table 1: Review of cases of syringocystadenoma combined with tubular apocrine adenoma

REFERENCE	AGE/GENDER	LOCATION	SEBACEOUS NEVUS
Toribio *et al.* (1987)	33/M	Scalp	Not mentioned
Ansai *et al.* (1989)	22/M	Scalp	Not mentioned
Epstein *et al.* (1990)	15/M	Mammary region	Not mentioned
Ishiko *et al.* (1993)	75/M	Scalp	Present
Aktepe *et al.* (2003)	19/M	Scalp	Present
Ahn *et al.* (2004)	52/M	Scalp	Present
Lee *et al.* (2005)	74/F	External auditory canal	Absent
Yamane *et al.* (2007)	77/F	Mammary region	Present
Vazmitel *et al.* 2008)	61/M	Scalp	Present
Kim *et al.* (2010)	40/F	Scalp	Present
Lee *et al.* (2011)	12/F	Lower dorsum	Absent
Yoon *et al.* (2011)	59/M	Calf	Absent
Present case (2014)	14/M	Dorsum	Absent

layer composed of columnar cells, where the characteristic decapitation secretion is observed, and an outer layer, formed by cuboidal cells. It also has a stroma with an infiltrate rich in inflammatory cells, mainly plasma cells. TAA has numerous irregular-shaped tubules with a dual cell layer. The tubules are embedded in a fibrous paucicellular stroma.[2-4] In the present case, the superficial portion of the tumor showed characteristics typical of SCAP and the deep portion was representative of TAA.

SCAP and TAA exhibit overlapping histopathology. Kazakov *et al.*[5] conducted an interobserver study for histopathological reassessment of TAA and SCAP performed by four dermatopathologists. The study confirmed the overlap between TAA and SCAP and demonstrated the absence of universally accepted diagnostic criteria for classifying lesions with morphological overlap between them, even among dermatopathologists and experienced pathologists.

Ishiko *et al.*[6] reviewed 19 cases of TAA described in the literature. In 10 of the 19 cases, the tumor was connected to the overlying epidermis, and it was necessary to differentiate the lesion from a SCAP. They described TAA as different from SCAP in some respects: TAA shows no cystic invaginations extending downwards from the epidermis, absent papillary projections and rare or absent plasmacyte infiltration.[7]

Our patient underwent excision of the lesion and has been followed at the service. ❑

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