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Tubular apocrine adenoma with syringocystadenoma papilliferum arising from the external auditory canal

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Abstract
Tubular apocrine adenoma (TAA) is a very rare sweat gland tumour. Comprehensive review of the literature reveals that TAA in the external auditory canal (EAC) has not yet been reported. We report a case of TAA in the EAC, together with characteristic histopathological findings.

Key words: External Auditory Canal; Adenoma; Apocrine Gland; Pathology

Introduction

A tumour occurring in the external auditory canal (EAC) is very rare. Exostosis is the most common benign tumour, and several benign adenoma may be found. In malignant cases, squamous cell carcinoma is most common, followed by basal cell carcinoma. Tubular apocrine adenoma (TAA) is a very rare sweat gland tumour. Since Landry and Winkelmann originally described this tumour as a new entity in 1972,[1] most cases have been reported at the scalp, extremity and eyelid.[2] However, TAA arising from the external auditory canal has not yet been reported. Recently TAA with syringocystadenoma papilliferum (SCAP) in the background of naevus sebaceous has been reported.[3] Here we report a rare case of TAA associated with SCAP, but without pre-existing naevus sebaceous in the EAC.

Case report

A 74-year-old woman was seen in consultation for a mass in the left EAC which she had had for 10 years. She had suffered from arthritis, hypertension, and mild hearing loss. She denied otalgia, ear fullness or tinnitus. The mass had recently increased in size and started to exude a foul-smelling serous discharge. The exudates had been present for two months and were unaccompanied by any other symptoms such as pain or hearing difficulty. Family history and laboratory findings were unremarkable. Histopathological diagnosis of a biopsy performed at a local clinic was basal cell carcinoma. We reviewed the biopsy slide and the diagnosis was revised as TAA. A re-biopsy was done and the diagnosis of TAA was confirmed. In the comprehensive otological evaluation, only mild sensorineural hearing loss was identified. There was no bony erosion in the EAC adjacent to the tumour seen in high resolution computed tomogram (HRCT). Examination revealed a 1.5 x 1 cm sized indurated tumour with central ulceration at the superoposterior EAC wall (Figure 1A). The tumour was completely excised under a surgical microscopic view (Figure 1B). For the denuded bone, we used split-thickness skin graft 0.25mm (10/1000 inch), packed with Merocel for rapid healing. The wound healed uneventfully, and there has been no evidence of recurrence for eight months.

Histopathology

The biopsy slide showed two different types of histopathological finding. In the upper part of the lesion, hyperplastic epidermis and epidermal invagination with papillomatous projections lined by two rows of epithelial cells were compatible with the diagnosis of SCAP (Figure 2). However, most tubules of deep dermis revealed a double layer pattern with hyalinized stroma. Although a focal basaloid pattern was noted in some tubules, the overall feature of the histology was not compatible with basal cell carcinoma. The surgical specimen was composed of circumscribed lobules of double-layered tubular structures situated in the deep dermis (Figure 3). The tubules had apocrine features with inner layer of cylindrical cells, often

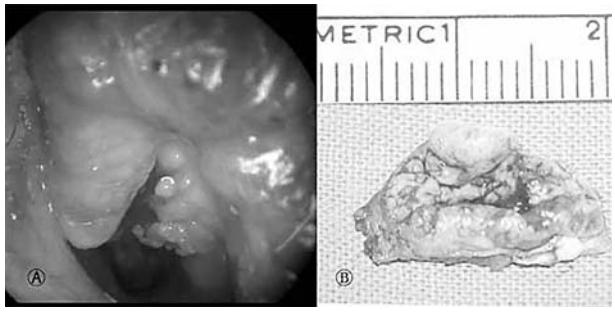

Fig. 1

(A) Otoscopic finding of the mass with central necrosis located at a superoposterior wall of the external auditory canal (EAC). The tympanic membrane is seen deep inside the EAC. (B) Surgical specimen with a scale bar: the 1.5 x 1 cm sized mass with central ulceration is shown.

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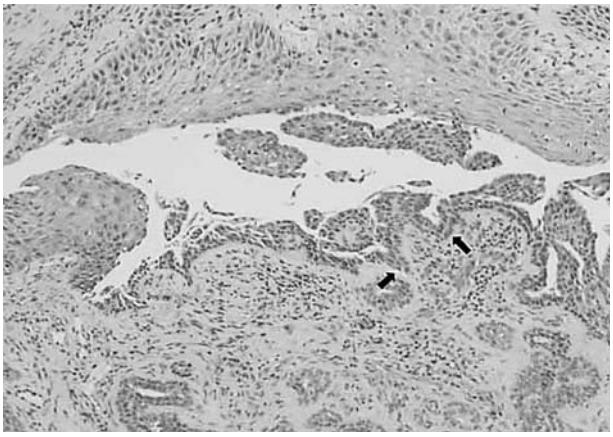

FIG. 2

The upper portion of the lesion shows epidermal invagination with papillomatous projection (black arrows), which is compatible with findings of syringocystadenoma papilliferum (H&E, x100).

showing decapitation secretion (Figure 4). Stromal invasion and cytologic atypia of tubules were not noted. The stroma consisted of hyalinized fibrous tissue with only a paucity of inflammatory cells. No papillomatous epidermal hyperplasia and mature sebaceous glands were identified. However, some ceruminous glands with lobulating contours could be found in the surrounding tissue. These findings in the deep dermis were compatible with the histopathological diagnosis of TAA.

- **This is the first reported case of a tubular apocrine adenoma occurring in the external auditory canal**
- **This tumour is a rare neoplasm of sweat gland origin**
- **Treatment was by complete surgical removal**

Discussion

In 1972, Landry and Winkelmann published the original description of TAA, whose histochemical and ultrastructural characteristics indicated its apocrine origin.[1] Umbert and Winkelmann suggested that TAA is an independent clinical entity consisting of a benign

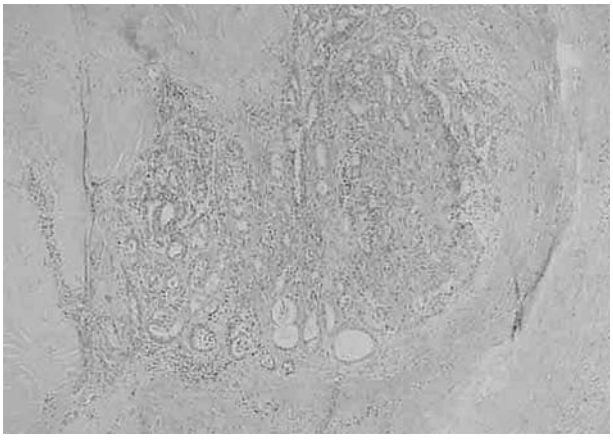

FIG. 3

The deeper portion of tubular apocrine adenoma shows double-layered tubular structures with lobulating contour (H&E, x40).

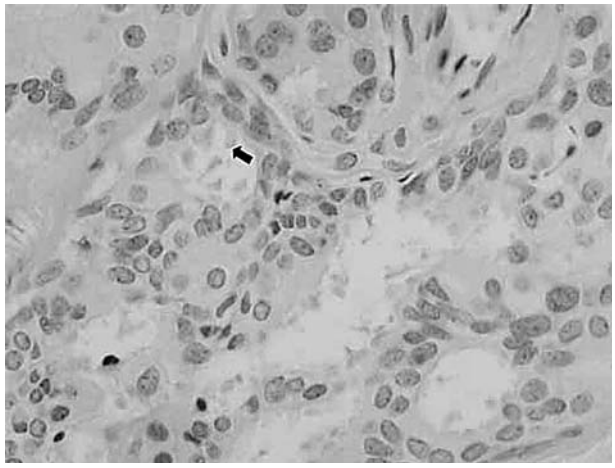

FIG. 4

Variable sized tubular structures of tubular apocrine adenoma are seen. Decapitation secretion of the luminal cell is also seen (black arrow) (H&E, x200).

appendage tumour of apocrine origin that is often associated with an organoid naevus.[4] Ishiko *et al.* reviewed 19 cases of TAA described in the literature.[5] Ten of the 19 lesions were located on the scalp and at least six cases involved a pre-existent organoid naevus. In most of the 19 cases, the tumour was connected to the overlying epidermis and it was necessary to differentiate the lesion from SCAP. They described TAA as being different from SCAP in several aspects: (a) TAA shows no cystically dilated apocrine invaginations extending down from the epidermis, (b) papillary projections are absent, and (c) infiltration of plasma cells is rare or absent. On the other hand, they suggested that some cases of TAA may arise from the pre-existing organoid naevus and that TAA and SCAP occur together in such cases. Therefore, TAA and SCAP might represent a spectrum of disease characterized by an appendage tumour of apocrine origin arising within an organoid naevus. Only five cases of TAA associated with SCAP have been reported in the literature and all of them had pre-existing naevus sebaceous.[6] However, TAA associated with SCAP without background naevus sebaceous (NS) has not yet been reported in any region of the body. Our case shows the characteristic findings of SCAP in the upper portion of the lesion and those of TAA in the deeper portion. However, no evidence of pre-existing naevus sebaceous could be identified in the biopsy and excision specimens. Only a small focus of ceruminous glands was noted in the deep dermis. Criber *et al.* have suggested that ectopic apocrine glands in naevus sebaceous may lead to both TAA and SCAP.[3] But our case was not accompanied with naevus sebaceous in surrounding tissue. The ceruminous glands of the surrounding tissue indicated the possibility of a histogenetic origin from apocrine cells of the ceruminous glands rather than from the ectopic apocrine glands in naevus sebaceous. Our case demonstrates that TAA with SCAP can arise without background nevus sebaceous in the non-scalp area. Although this current case was initially misdiagnosed as basal cell carcinoma, TAA are rarely combined with malignancy. Histologically, double-layered tubular structures without stromal invasion or cytologic atypia suggest a benign nature of the tumour. Only two cases of TAA have been reported to have malignant components.[4,7]

In summary, we present a rare case of TAA arising in the EAC associated with SCAP, but without background naevus sebaceous.

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Dr Yang-Sun Cho takes responsibility for the integrity of the content of the paper.
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