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Glandular Tumors of the External Auditory Canal*)

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ABSTRACT

We reported one case each of mixed tumor and adenoid cystic carcinoma originating in the external auditory canal, and one case of adenocarcinoma of the thyroid with the initial manifestation of symptoms in the otologic field eight years after the thyroidectomy. Major literature concerning ceruminous gland tumors were reviewed, and almost identical pathologic findings of ceruminous gland tumors to conventional sweat gland tumors, caution against metastatic cancers to the external auditory canal, the criteria of malignancy of ceruminous gland tumors and its unique biologic behavior were discussed.

INTRODUCTION

Haug[5] in 1894 reported a ceruminous adenoma and a case of what appeared to be adenoid cystic carcinoma as die Neubildungen des äusseren und mittleren Ohres. Since then, sporadic cases have appeared under various terminologies, such as ceruminoma, hidradenoma, cylindroma, myoepithelioma, mixed tumor, pleomorphic adenoma, clear cell carcinoma, tumors of ceruminous origin, adenomatous neoplasm, glandular tumor, cystadenoma, adenocarcinoma (or adenoma), ceruminous adenocarcinoma (or adenoma), carcinoid tumor, and adenoid cystic carcinoma. Because of relative rarity of glandular tumors in the external auditory canal (EAC), particularly in man as compared to animals, these tumors have escaped attention of not only otologisits but also pathologists, thus causing much confusion of the terminology. We present two cases of the ceruminous gland tumors originating in EAC, and one case of adenocarcinoma of the thyroid unusually with the intial manifestation of symptoms in the otologic field eight years after the thyroidectomy. The third case mimicked histologically adenoma or adenocarcinoma of EAC and only necropsy confirmed its metastatic origin. These three cases were experienced in recent six-year period.

CASE PRESENTATION

Case 1: This is a 52-year-old male complaining of the left ear obstruction of two to three months' duration. A tumor was situated about 0.5 to 1.5 cm from the entrance of the left EAC by attaching to the posterior wall. Through an endoaural incision, it was easily removed; the tympanic membrane was intact. The tumor was smooth on the surface, elastic firm, spherical, measured approximately one cm in diameter and weighed 0.5 g.

Case 2: This is a 59-year-old male, complaining of a tumor of the right EAC for 10 years, hearing disturbance for two years and aural bleeding for one to two months. A tumor was situated at the cartilaginous portion of the right EAC by attaching to the cranial wall; tympanic membrane was intact. Under the diagnosis of a malignant tumor, 3,000 rads (10 MeV electron beams) of preoperative irradiation was given. The tumor together with the cartilaginous portion of EAC was removed *en block*, followed by local skin grafting. It was

*) 田仲俊雄，斉藤龍介，石原基正，大道卓也，小倉義郎：外耳道に発生した腺腫および腺癌

pinkish, smooth on the surface although hemorrhagic due to erosion, spherical, and measured approximately 1.5 cm in diameter.

Case 3: This is a female born in January 1920. In 1936, she had radiotherapy for hyperthyroidism once a month for about one year. In 1966, she noticed a foreneck tumor. Because of its gradual enlargement, in August 1968, she consulted the Surgical Department, Okayama University Hospital, and under the diagnosis of a carcinoma she had the left radical neck dissection including the left hemithyroidectomy, followed by Co^{60} therapy. About eight years later, in September 1976, she started obstructive sensation in the left ear. Because of its worsening associated with hearing disturbance, in September 1977, she visited the Otolaryngology Department, Okayama University Hospital, and had the left radical tympanomastoidectomy including the sleeve resection of entire EAC in November. A friable, easily bleeding, granulomatous tumor occupied the osseous portion of EAC and entire middle ear involving the auditory ossicles; the originating site of the tumor was not certain due to extensive tumor infiltration. Postoperatively, she received 5,600 rads of Linac irradiation, and was discharged in March 1978. In January 1979, the tumor around the left sub- and retroauricular regions enlarged to child's head size together with swelling of the left soft palate due to the left parapharyngeal infiltration, and the left accessory and hypoglossal paresis. In October, chest x-rays disclosed a coin lesion of 1.0×1.5 cm in the right upper field; this gradually enlarged. In May 1980, tumorous swelling extended to the right pharynx to necessitate tracheostomy. The patient died in March 1981. Necropsy proved a follicular adenocarcinoma of the thyroid metastasizing to the left sigmoid sinus, cerebellum and left auditory canal, which extended to the parotid, submandibular region and pharynx of the same side, further deep to the left subclavicular vein and para-esophageal area, and to bilateral lungs as small metastatic foci.

PATHOLOGIC FINDINGS

According to the study made by several investigators[2,8,9,12], special stainings employed for these three cases included periodic acid-Schiff (PAS), PAS with diastase digestion, mucicarmine, alcian blue, prussian blue, Ziehl-Nielssen, Sudan black after paraffin embedding and Masson's trichrome.

Case 1: The tumor was slightly knobby on the surface and encapsulated by thin fibrous strands (Fig. 1). There were relatively well-differentiated tubular structures with flat myoepithelial cells (Fig. 2), cartilaginous area stained deeply with alcian blue (Fig. 3), epithelial strands surrounded by myxoid substance (Fig. 4), well-differentiated squamous cell clusters (Fig. 5), and a few calcified foci. The diagnosis was a mixed tumor, probably benign.

Case 2: The tumor obviously infiltrated to the surroundings without clear-cut capsule, and had an ulceration with hemorrhage on the EAC surface connecting to the tumor mass (Fig. 6). The tumor cells were widely separated by thick hyaline substance of PAS-positivity and diastase-resistance (Fig. 7). There were duct-lining cells surrounded by myoepithelial cells with vacuolated cytoplasm (Fig. 8). In some areas, tumors were composed only of apparent myoepithelial cells containing faintly PAS-positive substance (Fig. 9). Only the periphery of the main tumor showed a few foci consisting of tubular, duct-like structures (Fig. 10). The diagnosis was an adenoid cystic carcinoma.

Case 3: The initial biopsy taken from a nodule attached to the tympanic membrane consisted of two pieces of tiny nodules surrounded by squamous cell layer (Fig. 11). It had relatively well-differentiated tubular structures with mild nuclear pleomorphism and without mitosis (Fig. 12); a possible ceruminous gland adenoma was made. The second materials by the radical surgery, consisting of several specimens taken from the EAC and middle ear, were essentially similar to the first biopsy. But, because of extensive tumor infiltration clinically, the diagnosis was altered to a possible ceruminous adenocarcinoma. Afterwards, the specimens obtained from thyroidectomy and necropsy were examined retrospectively; these were essentially identical to the specimen of the initial biopsy.

DISCUSSION

According to the survey of Conley and Schuller[3] between from 1945 to 1972, malignant EAC tumors include squamous cell carcinoma

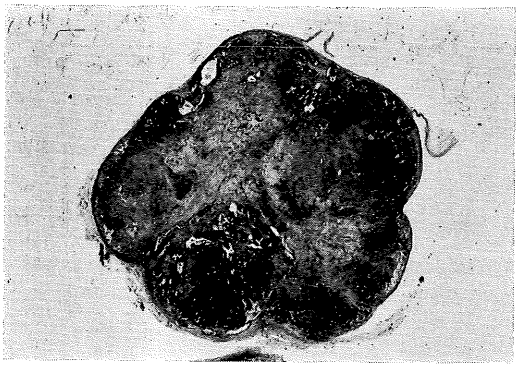

Fig. 1. Panorama view of the excised tumor encapsulated by thin fibrous strands (Case 1). Masson's trichrome.

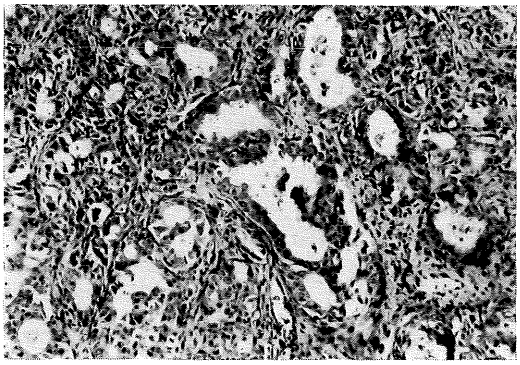

Fig. 2. Relatively well-differentiated tubular structures with flat myoepithelial cells (Case 1). Masson's trichrome, ×200.

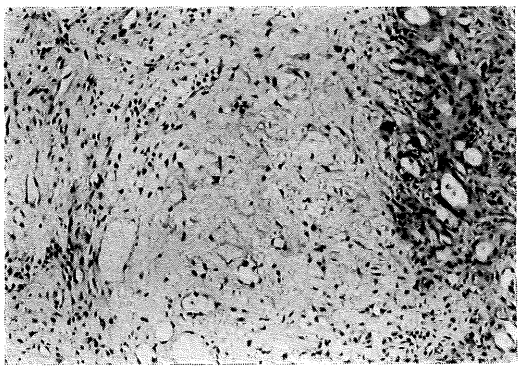

Fig. 3. Cartilaginous area adjacent to tubular structures (Case 1). H. E., ×100.

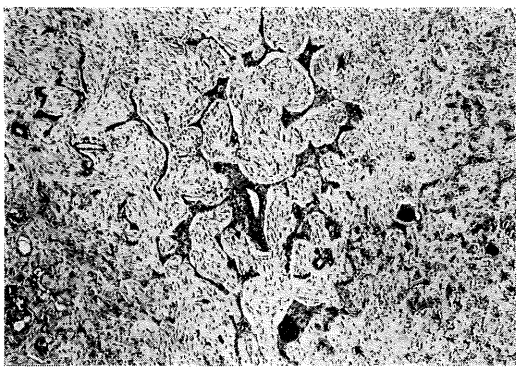

Fig. 4. Epithelioid strands surrounded by myxoid substance (Case 1). H. E., ×40.

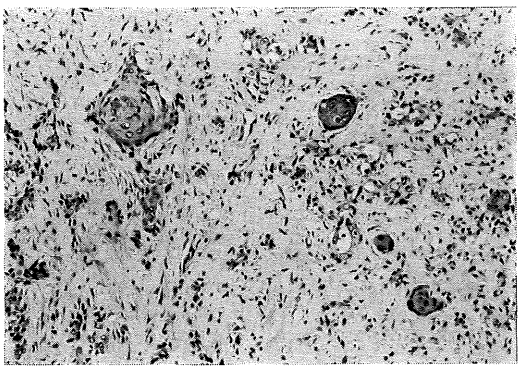

Fig. 5. Several well-differentiated squamous cell clusters (Case 1). H. E., ×100.

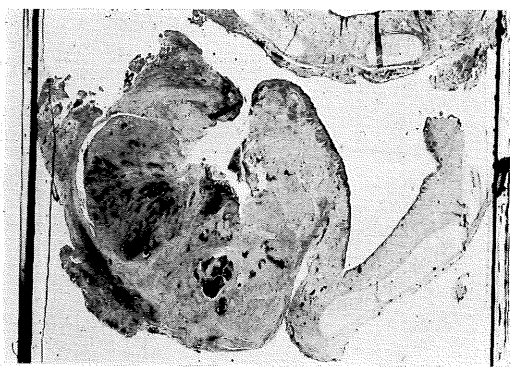

Fig. 6. Panorama view of the excised tumor infiltrating to the surroundings without clear-cut capsule, and with ulceration, reaching the tumor mass, on the surface of external auditory canal (Case 2). Masson's trichrome.

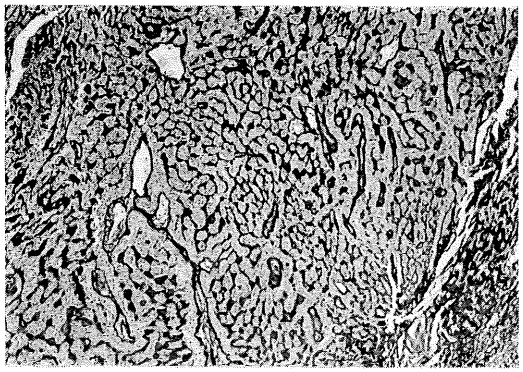

Fig. 7. Tumor cells separated by thick, PAS-positive, hyaline substance (Case 2). PAS, ×40.

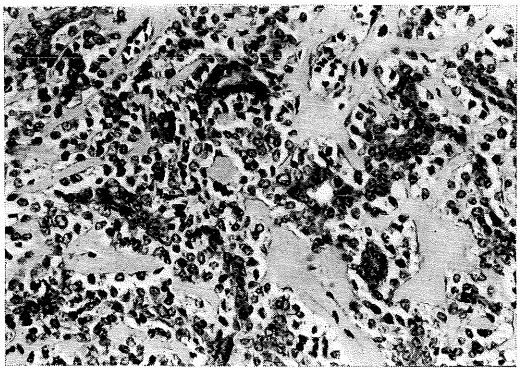

Fig. 8. Duct-lining cells surrounded by myoepithelial cells with vacuolated cytoplasm (Case 2). H. E., ×200.

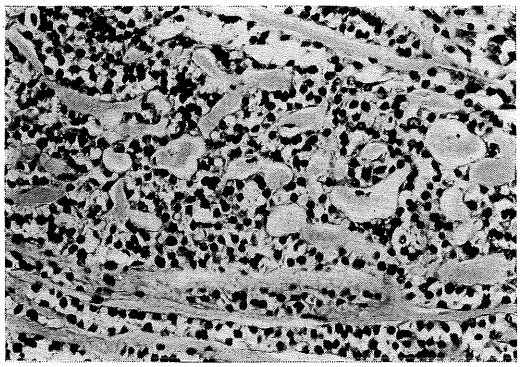

Fig. 9. Groups of myoepithelial cells containing faintly PAS-positive substance (Case 2). PAS, ×200.

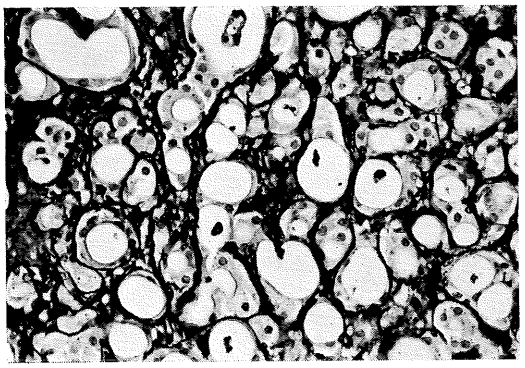

Fig. 10. Well-differentiated, tubular, duct-like structures (Case 2). PAS, ×200.

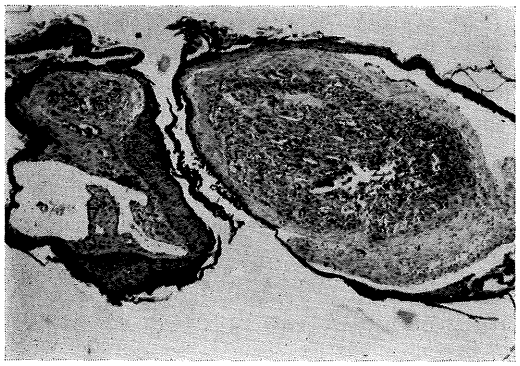

Fig. 11. Panorama view of the initial biopsy specimen, consisting of two tiny nodules surrounded by squamous cell layer (Case 3). H. E., ×40.

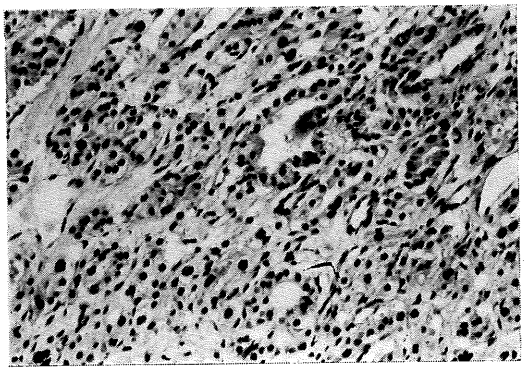

Fig. 12. Relatively well-differentiated tubular structures with mild nuclear pleomorphism (Case 3). H. E., ×200.

59%, adenocarcinoma 9.8%, adenoid cystic carcinoma 9.8%, basal cell carcinoma 8.2%, malignant melanoma 4.9%, paraganglioma 1.6% and others 6.6%, indicating over half of squamous cell carcinoma and about 20% of the glandular origin. Canker and Crowley[2] in 1964 reported seven cases consisting of four cases with adenoma of ceruminous gland type, and one each of ceruminous gland carcinoma, carcinoma of cylindromatous type and mixed tumor of EAC. Reviewing world-wide literature, Wetli et al.[2] in 1972 produced a simplified classification of benign and malignant EAC tumors derived from the ceruminous glands, *i.e.*, adenoma (Ad.), adenocarcinoma (Ad. ca.), adenoid cystic carcinoma (Ad. cy. ca.), and mixed tumor (Mix. t.). Pahor and O'Hara[9] in 1975 made a comprehensive survey which consisted of 14 Ad., 7 Ad. ca., 38 Ad. cy. ca., and 7 Mix. t. including their own-experienced one case with Mix. t. Hicks[6] in 1983 comprehended 21 Ad., 20 Ad. ca., 55 Ad. cy. ca. and 6 Mix. t. including their own-experienced 4, 2, 3 and 1 cases, respectively. Hicks' series included also 37 cases reported by Pulec[11] in 1977, especially Ad. cy. ca.

Our three patients consulted the Otolaryngology Department because of lumps visualized in EAC. The lumps were situated external to the tympanic membrane, except for Case 3 which extended to the middle ear. Therefore, tumors of Cases 1 and 2 were originated definitely from glandular structures of the EAC. The normal EAC contains both sebaceous and ceruminous glands. The latter is a modified sweat gland, which is composed exclusively of ceruminous apocrine glands. Although no eccrine glands are present in EAC, tumors derived from ceruminous glands are histologically identical to sweat gland tumors elsewhere. Our Case 1 demonstrated chondroid matrix with alcian blue, and Case 2 stained hyaline stroma deeply with PAS, as seen in sweat gland tumors. Other special stainings were also not particularly helpful to distinguish the sweat gland tumor from ceruminous gland tumor. Ham and Cormack[4] emphasized that "the names *apocrine* and *eccrine* used in connection with sweat glands can suggest inappropriate connotations about the ways their secretions are released", and that "these two are rightly classed as merocrine".

Concerning the histologic similarity of adenocarcinoma of the thyroid to that of the ceruminous glands, Pallanch et al.[10] observed an atypical glandular tumor in their Case 9 out of the 11 cases with adenocarcinoma and adenoma of the middle ear. This particular tumor had persistent recurrence 23 years after the onset of symptoms and histologically "somewhat resembles thyroid cancer", although the thyroid gland was normal. In our Case 3, i) the initial biopsy obtained from a nodule from the periphery of the typmanic membrane resembled a ceruminous adenoma, at least histologically. Furthermore, ii) specimens by biopsy and radical surgery performed in the Otolaryngology Department did not contain colloid substance in glandular lumens, as often seen in thyroid carcinoma. iii) Pathology report at the time of thyroidectomy was simply "undifferentiated carcinoma" without any further histologic details. In addition, iv) we thought the recurrence of thyroid cancer over eight years and its initial metastasis to the EAC to be rather unlikely. These several reasons misled us as to the interpretation of the initial biopsy specimen.

Regarding the malignancy of glandular neoplasm of EAC, Batsakis et al.[1] reported two cases of so-called ceruminoma with extremely different pattern of aggressiveness even though histologically they looked almost identical. They concluded that "their biological activity cannot be predicted by the histological appearance which is often benign". Adenoid cystic carcinoma of EAC, which corresponds to our Case 2, has been known to manifest a high incidence of local recurrence and systemic spread.[6] On the other hand, our Case 3 presented with reasonably well-differentiated tubular pattern; the biological activity of EAC tumors can be sometimes quite unpredictable. Therefore, we would like to draw attention to the criteria of the malignancy which was set tentatively by Pallanch et al.[10] in glandular tumors of the middle ear, that is, the presence or absence of mitoses and cellular pleomorphisms (histologically), and invasiveness to adjacent bone, soft tissue, perineural space or cranial nerves and recurrence of tumor within 10 years (clinically). The problem of the recurrence within 5-year period was also emphasized by Hicks[6] in glandular tumors of EAC.

Johnstone et al.[7] in 1957 studied five cases

of hidradenoma (or so-called ceruminoma) of their own experience and 10 previously reported cases of adequate description. They concluded that the EAC hidradenoma is often malignant in contrast to that seen in the general skin surface; the latter is less than 5% in malignancy. Ceruminous gland tumors of EAC should be regarded as a unique disease entity, particularly in their biological behavior which certainly relates to appropriate choice of treatments.

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