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Papillary Carcinoma Arising from a Thyroglossal Cyst —A Case Report—

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ABSTRACT. A number of congenital lesions as well as malignant may arise in the neck. Thyroglossal cysts and sinuses are the most common anomalies encountered. They are derived from vestigues of the thyroglossal duct connecting the primitive median thyroid to its point of origin on the ventral wall of the pharynx. The presence of ectopic tissue in the thyroglossal cyst is occasionally observed. However, papillary carcinoma arising from a thyroid remnant is uncommon. In this paper, a case of papillary carcinoma developing from a thyroglossal cyst in a housewife of 47 was presented and its etiology was discussed.

CASE REPORT

A 47-year-old housewife was referred to our clinic for further investigation of a lump in the neck in June 1985. The lump had been noted at the beginning of June. It had increased in size incidiously without pain. On physical examination, a round smooth cystic mass, measuring 4.4 cm in diameter was found on the midline of the upper anterior portion of the neck. It was displaceable and did not adhere to the overlying skin but it was fixed to the hyoid bone. There was no goiter or cervical lymph node enlargement.

Routine laboratory examinations were within normal limits. A thyroid scintigram with ^{99m}Tc pertechnetate delineated both lobes to be of normal size, shape, and configuration, but no accumulation was seen in the tumor area. Soft tissue x-rays of the neck failed to demonstrate any abnormal calcification. Ultrasonogram revealed a cystic pattern that included papillary protrusion in part in the corresponding area.

At surgery, a cystic mass measuring 4×3 cm was found lying immediately beneath the sternohyoid muscle. There were no adhesion or inflammatory reaction to surrounding tissues. The cyst was connected to a duct which was found passing above the hyoid bone. The cyst and duct were separated from

芝田　努，原田種一，片桐　誠，大久保茂樹，川野　亮，崔　哲旬，岡村泰彦，三宅一昌，一本杉聡，山根康彦，大浜寿博，園尾博司，妹尾亘明

the surrounding tissues and the duct was removed up to the proximal portion of foraman cecum of the tongue. The thyroid gland was intact and there was no enlargement of the cervical lymph nodes.

Glossly, the specimen consisted of an ovoid piece of tissue which was cystic and measured 4 cm in diameter.

Although most of the inner surface of the cyst was smooth and whitish gray in color, there was an area slightly protruding into the lumen with reddish solid tissue measuring 1.2 cm at the greatest dimension (Fig. 1). Microscopically,

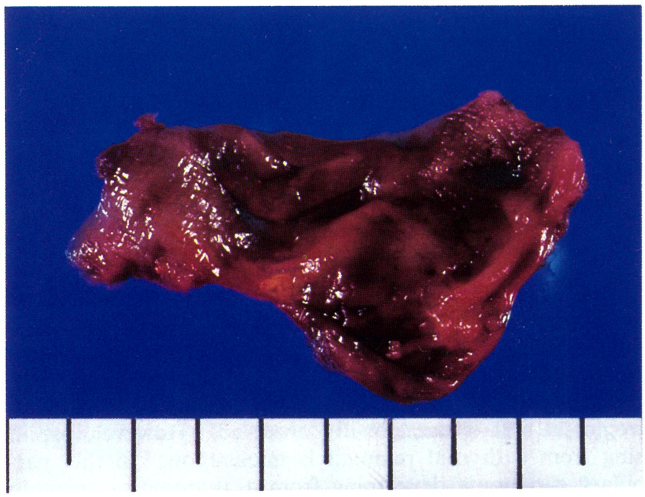

Fig. 1. Inner surface of the thyroglossal duct cyst including a slightly protruding area.

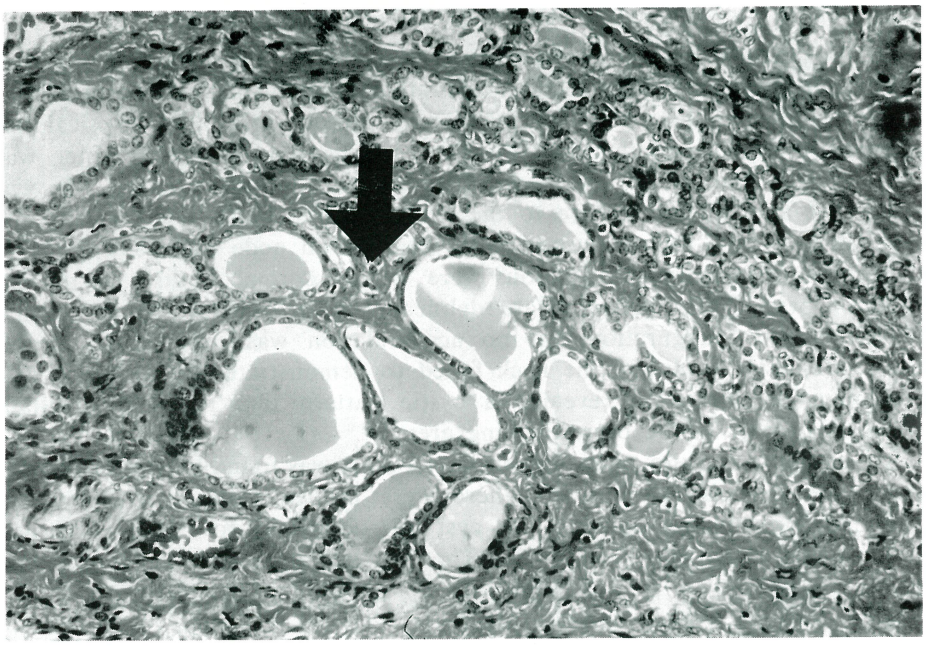

Fig. 2. Thyroid follicles in fibrous connective tissue in the cyst wall indicated by arrow.

the cyst wall consisted of fibrous connective tissue without recognizable internal lining epithelium and contained several islets of thyroid tissue (Fig. 2). In addition, a feature of papillary carcinoma was proved in the grossly protruding solid area and neighbouring islets in the wall (Fig. 3). Psammoma body-like

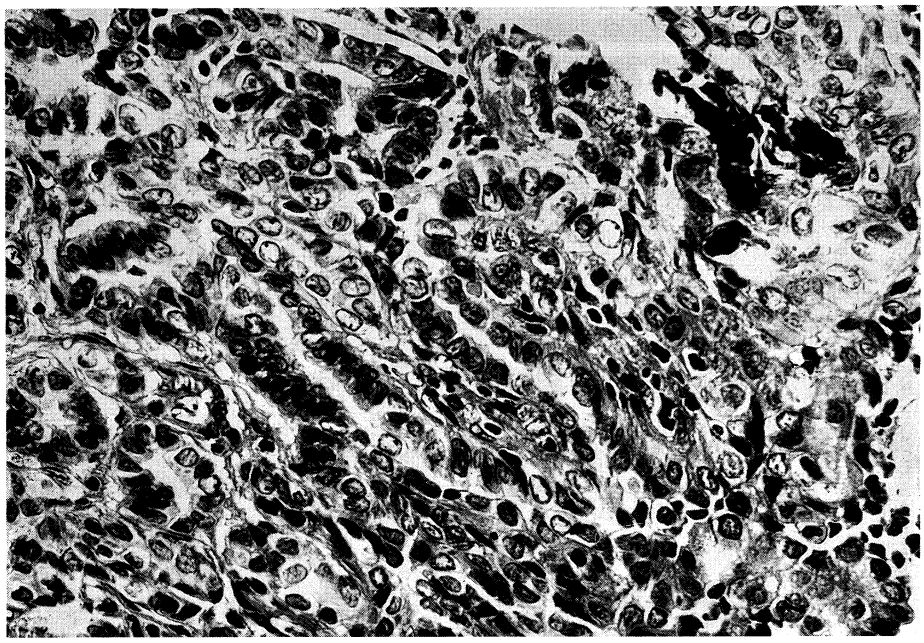

Fig. 3. A focus of papillary carcinoma within the cyst wall.

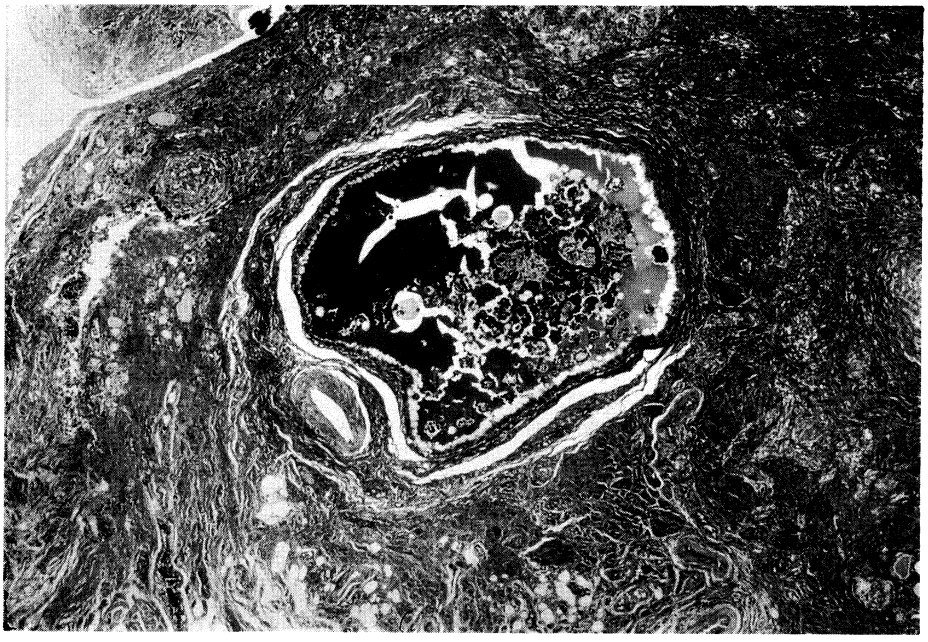

Fig. 4. Another focus of papillary carcinoma showing fine calcifications simulating psammoma bodies.

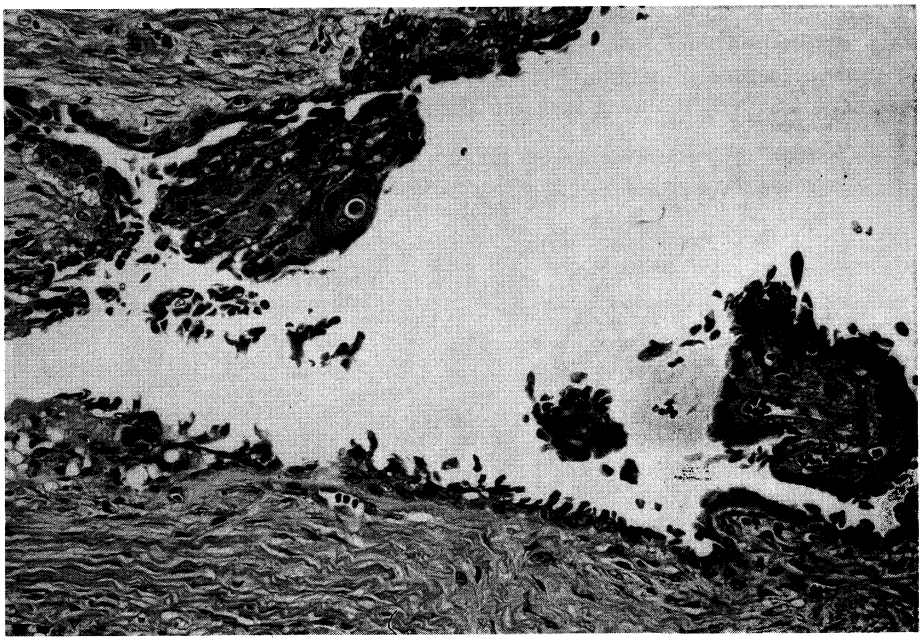

Fig. 5. An area of squamous metaplasia in papillary carcinoma exhibiting pearl formation.

calcifications and squamous metaplasia were also seen (Figs. 4, 5).

The patient is clinically free of disease 6 months following the surgery. She is required to return to the outpatient clinic twice a year for observation.

DISCUSSION

The thyroglossal duct is a vestigial structure which passes from the foraman cecum at the base of the tongue to the isthmus of the thyroid gland. If a portion of the duct remains unclosed, a cyst lined with columnar, squamous or transitional epithelia can form.[1)]

With the embryological development mentioned above, a thyroglossal cyst containing normal thyroid tissue in the wall is not uncommon. According to the report collected by Pollock[2)] and Ward,[3)] the presence of histologically identifiable thyroid epithelium in the midline thyroglossal remnant was between 5% and 45%.

However, the incidence of malignancies arising from thyroid tissue in the thyroglossal tract is rare and they have been reported in less than 1% of all cases.[5)]

According to Hanamatsu,[6)] 88.6% of these malignancies were as papillary carcinoma, 5.5% as squamous carcinoma, 1.8% adenocarcinoma, 0.9% follicular carcinoma and 0.9% as anaplastic carcinoma. Thyroglossal malignancies other than thyroid carcinoma were thought to be extremely rare, although a few cases of epidermoid carcinoma were reported. Jaques *et al.*[1)] were not able to find a satisfactory case of a tumor confirmed to have arisen from the epithelium of a thyroglossal cyst wall in the files of Armed Forces Institute of Pathology.

Carcinoma of a thyroglossal cyst occurs more frequently in women and the age of the patients at the time of diagnosis ranged from 6 to 81.[1] The peak incidence for females is in the forth decade and for males in sixth decade.[4]

When a papillary carcinoma is encountered in a thyroglossal cyst, the thyroid gland must be always considered to be incriminated as the source.[7]

In the present case, neither the thyroid gland itself nor any mass in the thyroid were palpated. A scintigram delineated a normal thyroid configuration without any defects.

As for the management of papillary carcinoma arising from a thyroglossal cyst, several alternative methods exist. In most cases, the possibility of malignancy is not suspected prior to surgery. Therefore, the Sistrunk procedure[8] or extirpation of the cyst with high ligation and removal of its duct might be performed. Whether additional surgery is necessary or not has been debated, since optimal treatment of papillary carcinoma of the thyroid gland itself remains controversial. We believe that further surgical interventions are not indicated since papillary carcinoma of thyroid gland is an indolent disease and neck dissection is not always necessary, although follow-up observation is mandatory.

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