A Case Report of Papillary Carcinoma Arising from a Thyroglossal Duct Cyst

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An anterior neck nodule was found in a 32-year-old female patient. Physical examination showed a 2.5 cm soft nodule that overlay the upper mid-part of the thyroid cartilage and moved during swallowing. Ultrasonography with fine needle aspiration (FNA) was performed preoperatively. No solid component was found to exist within the cyst. No other nodules were found to exist within the thyroid gland. The FNA cytology was negative for malignancy. The patient only underwent complete excision of the cyst. Postoperatively, the histopathological study revealed primary papillary carcinoma within a thyroglossal duct cyst. Radioiodine scanning revealed no cold nodules. Serum thyroglobulin was below the reference range. The patient received follow-up studies as an outpatient for six months. *(Chang Gung Med J 2006;29(4 Suppl):13-6)*

Key words: papillary carcinoma, thyroglossal duct cyst, thyroid neoplasm, aspiration cytology.

The thyroid gland develops to form a median diverticulum between the first and second pharyngeal pouches, and immediately dorsal to the aortic sac at the 4th week of fetal life. The connection of the median diverticulum with the pharynx is termed the thyroglossal duct. From the foramen cecum, it extends caudally in the median line ventral to the hyoid bone. The distal part of the duct commonly differentiates variably as the pyramidal lobe and suspensory fibrous band of the thyroid gland late in fetal life. Occasionally parts of the thyroglossal duct persist and form aberrant masses of thyroid tissue cysts, fistula or sinuses, usually in the midline.(1) Carcinoma is found in 1% to 2% of thyroglossal duct cysts (TDC).(2) The average age at diagnosis is 39.3 years (range 22 to 68 years).(3) Eighty five percent of TDC carcinomas are of the papillary type.(4)

In this report, papillary carcinoma grew in a midline neck cyst. It is presented with regard to the histopathological findings.

CASE REPORT

An anterior neck nodule was found in a 32-year-old female patient two years previously. She had no other evidence of disease. The nodule did not cause any discomfort in her daily life. She visited the Chang Gung Memorial Hospital because the nodule had gradually increased in size. On physical examination, there was a 2.5 cm soft nodule that overlay the upper mid-part of the thyroid cartilage and moved during swallowing. The thyroid gland and lymph nodes in bilateral jugular chains remained normal. Routine laboratory examinations and thyroid function tests were within the reference ranges. Ultrasonography (US) with fine-needle aspiration (FNA) was done. The US revealed an anechoic cyst. No solid component existed within the cyst. The FNA cytology showed macrophages and old red blood cells in a thick colloid background, and was negative for cancer. A repeat US with FNA per-

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Received: Nov. 30, 2004; Accepted: May 13, 2005
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formed four months later was also negative for malignancy.

The patient was admitted with the presumptive diagnosis of a benign TDC. At the time of surgery, the cyst had no parts connecting it to the thyroid gland. It was removed along with the tract extending to the mid-portion of the hyoid bone. There was no lymphadenopathy.

In the postoperative histopathological study, gross examination of the specimen showed a well defined cystic mass, which measured 3.6 x 2.7 x 2.5 cm. Microscopic examination revealed extensive cystic change and chronic inflammatory changes within the cyst. No normal ectopic thyroid tissue was identified in the entire cyst. The wall of the cyst was composed of fibrous stroma without epithelial lining. One papillary tumor was attached to a small portion of the cyst wall that measured < 1 cm in diameter. The carcinoma cells had grooved and ground glass nuclei. The tumor was confined within the wall of fibrous stroma. (Fig. 1) No lymph node tissue was found in the cyst. One month after surgery, T3, T4, thyroid stimulating hormone (TSH) levels and radioiodine scanning revealed no abnormalities. Four months after surgery, repeat I^{131} thyroid scanning revealed no abnormality. Thyroglobulin serum level six months after surgery was below the reference range.

DISCUSSION

The findings for papillary carcinoma, by FNA, in a TDC include high cellularity, papillary structure, and cells with enlarged nuclei that have nuclear grooves and intranuclear peudoinclusions.[5,6] These diagnostic criteria for a papillary carcinoma in a TDC are the same as in the thyroid gland. However, reaching a preoperative accurate diagnosis of papillary carcinoma in a TDC by FNA is uncommon: the true-positive rate was 53% and the true-negative rate was 47%.[5] In this report, the patient received US with FNA twice. The FNA cytology failed to reveal malignancy. The reason was that a small-sized cyst with an occult papillary carcinoma caused false-negative results.

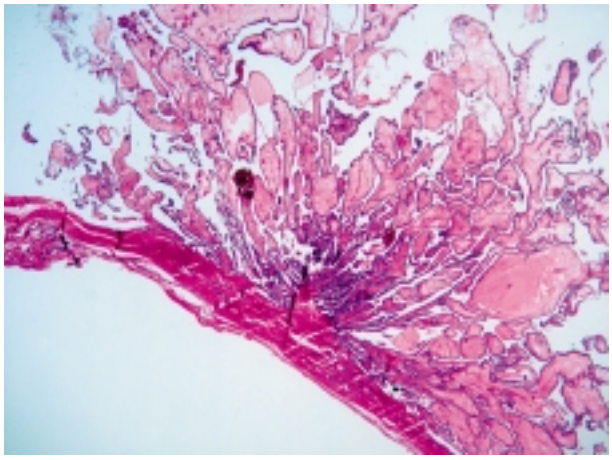

Fig. 1 A papillary carcinoma was attached to the inner cyst wall. (H&E stain, x 4)

In our case, histopathological study revealed absence of an epithelial lining and normal thyroid follicles in the wall of the cyst. Nevertheless, the cyst in the midline extended along with the tract to the mid-portion of the hyoid bone. No lymphoid structure existed in the TDC. These findings suggested the diagnosis of primary papillary carcinoma arising from a thyroglossal duct remnant.[7]

The indications for surgery on a TDC include increasing size, cyst infection and presence of carcinoma. The acceptable treatment of TDC carcinoma is the Sistrunk procedure.[2] The reported cure rate associated with this procedure is 95%.[8] In this report, the patient received only excision of the cyst. Was the treatment for her pertinent? Did she need further radical surgery?

In the literature, M. O'Connell et al. reported on eight cases of TDC carcinomas with mean follow-up of 16.5 years (range six to 41 years).[9] Four cases with papillary carcinoma received limited excision only. The survival time without recurrence was more than 10 years. The study recommended a more conservative approach if the tumor was small and better prognosis of differentiated thyroid carcinoma in a young population, especially if the patients are female.[9]

In our case, this patient had no nodules in the thyroid gland, no previous exposure to radiation, no lymph node metastases and no extracapsular spread of the TDC carcinoma. She received only complete excision. She needs further life-long follow-up with physical examinations and imaging studies.

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乳頭狀上皮癌存在於甲狀舌咽管囊腫

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一位 32 歲女性病人發現頸部前有一結節，物理學檢查顯示：在甲狀軟骨上部的中間有一 2.5 公分的囊腫，它會伴隨吞嚥而移動，她接受超音波及細針抽吸術檢查，超音波顯示在無其他的結節存在於甲狀腺中，細胞學結果顯示無惡性細胞的存在。然後，他接受囊腫切除手術，術後病理學報告發現：在囊腫內有一乳頭狀上皮癌，術後，放射性碘甲狀腺掃描攝影顯示沒有異常，血清中甲狀腺球蛋白在正常範圍內，截至今爲止，她共接受 6 個月的門診追蹤；本文將討論這個罕見病例的診斷及處理方式。(長庚醫誌 2006;29(4 Suppl):13-6)

關鍵字：乳頭狀上皮癌，甲狀舌咽管囊腫，甲狀腺腫瘤，細針抽吸術。

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受文日期：民國93年11月30日；接受刊載：民國94年5月13日

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