



Invasive and Metastasis Primary Papillary Carcinoma Arising in Thyroglossal Duct Cyst: A Case Report

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Abstract

Background: Thyroglossal duct cyst is the most common developmental anomaly of the thyroid gland, while thyroglossal carcinoma with local invasion and cervical metastasis is particularly rare.

Case Summary: We present a female patient with parathyroid mass and swollen lymph diagnosed as papillary thyroid carcinoma with cervical metastasis. The 29-year-old patient was referred to our hospital due to a solid-cystic swelling in the submental region adjacent to the hyoid bone, which was not in conformity to a thyroglossal duct cyst as the solid part enhanced directly. Through the Fine-Needle Aspiration Cytology (FNAC) evaluation from the tumor and lymph nodes, a diagnosis of papillary carcinoma and lymph nodes metastasis was made. The patient was subjected to a Sistrunk's procedure, bilateral thyroidectomy, as well as bilateral cervical dissection.

Conclusion: In such patients, the proper treatment protocols should be carried out after a definite diagnosis through complete preoperative examinations, involving computerized tomography, magnetic resonance imaging, thyroid scintigraphy, and thin-needle aspiration biopsy particularly.

Keywords: Thyroglossal duct cyst; Papillary carcinoma; FNAC; Metastasis; Sistrunk's operation; Thyroidectomy

Introduction

The incidence of papillary carcinoma arising in thyroglossal duct cyst is rare, and often misdiagnosed preoperatively as thyroglossal duct cyst for clinical rarity. And the diagnosis usually depends upon postoperative pathological examination by incidence. Local invasion and regional lymph node metastasis of thyroglossal duct cyst carcinoma rarely occurs of reported cases. Hence, careful preoperative examinations of these cysts are necessary, and operation plan should be formulated comprehensively.

In this paper, we report a case of primary papillary carcinoma occurring in a thyroglossal duct cyst with local invasion and multiple cervical lymph node metastasis.

Case Presentation and Results

A 29-year-old female presented to the Ear-Nose-Throat (ENT) department with a history of midline neck swelling of half year duration. She did not complain of sore throat, dysphagia, and hoarseness, neither history of neck surgery or radiation. On physical examination, an irregular, painless, obscure boundary, fluctuant, 4 cm × 2 cm mass was in the middle of the neck at hyoid bone level, and it showed vertical movement on deglutition. She was initially diagnosed with thyroglossal duct cyst. In the laboratory findings, her Free Triiodothyronine 3 (FT3), Free Triiodothyronine 4 (FT4), and Thyroid Stimulating Hormone (TSH) were normal. Ultrasonography showed no nodules in both thyroid lobes. On a thyroid scan, the whole thyroid showed decreased uptake, while the cyst did not show any uptake of ^{99m}TcO₄, without cold spots in the thyroid. CT (Figure 1A, 1B) and MRI (Figure 1C, 1D) examination showed a multinodular lesion adjacent to hyoid bone, and the solid components obviously strengthened and burrs (Figure 1A, 1B, 1D) were observed on the edges after enhancement, which suggested a malignancy tumor possibly. Besides, multiple lymph nodes enlarged and enhanced bilateral neck, of which the biggest one was 1cm in diameter. Then she was referred to stomatology department considered with submaxillary type sublingual gland cyst. For further diagnosis, the patient was subsequently referred for FNAC.

In the FNAC evaluation, the thyrocytes with uniform nuclei made up single-layer groups was observed, which conformed to the feature of papillary carcinoma of thyroid gland (Figure 2A). Considering of multiple enlarged cervical lymph nodes, the patient was then referred to lymph

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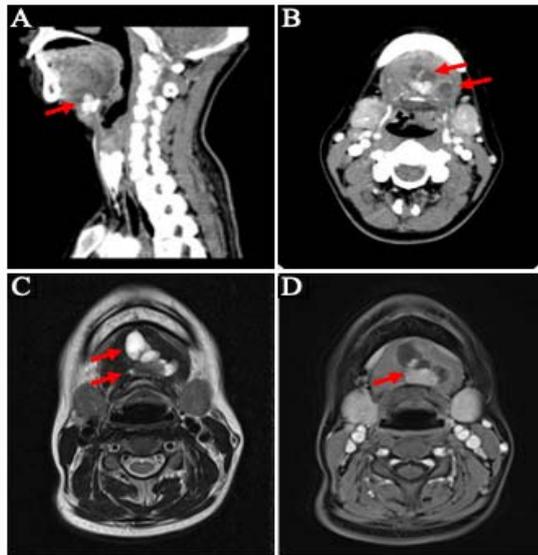


Figure 1: Computed Tomography (CT) and magnetic resonance imaging of the mass. a 4 cm × 2 cm diameter multilocular, midline, thyroglossal duct cyst was confirmed (red arrow), with obviously strengthened solid components and burrs on the edges after enhancement. A and B showed CT scan; C and D represented MR scan.

nodes biopsy, and the same results were obtained with the tumor (Figure 2B). Finally, a diagnosis of papillary carcinoma of thyroglossal duct cyst with local invasion and cervical metastasis was made and the patient was transferred to the department of thyroid and breast surgery for further therapy. She received an operation of total thyroidectomy, bilateral modified radical neck dissection, pyramidal lobe excision, and a conventional Sistrunk's operation.

Tissues were sent for histopathological examination. Gross examination showed a mass measuring 2.5 cm × 2.5 cm, with an attached part of hyoid bone and skeletal muscle. The final diagnosis of primary papillary carcinoma in thyroglossal duct cyst was confirmed (Figure 3A). Moreover, the patient had tumor's local invasion and multiple nodal metastasis (Figure 3B), including central zone (2/15), left cervical region (1/14), right cervical region (3/14), and parathyroid region (4/5), while no malignant cell was found on thyroid gland and adjacent tissue. She received postoperative thyroid hormone therapy and regular follow-up showed no recurrence and other abnormality.

Discussion

Thyroglossal duct cysts are the most common developmental anomalies, which usually locate in the midline or, less frequently, in the lateral anterior region of the neck, usually with no clinical symptom [1]. Carcinoma arising in the thyroglossal duct cyst is pretty rare, only accounting for 1% of all cases and seen often in younger women [2,3]. The cause of thyroglossal duct cyst carcinoma is unclear. The predominating theories are either metastatic disease from an occult primary or spontaneous development from ectopic thyroid tissue, while some cases are considered as metastatic thyroid carcinoma [4]. Papillary carcinoma is by far the most common type (75% to 80%), but other types of primary tumor do occur, such as mixed papillary-follicular carcinomas, squamous cell carcinoma, and follicular carcinoma [5].

The diagnosis of thyroglossal duct cyst carcinoma is frequently made postoperatively by histopathological examination of the resected tissues, which may be clinically indistinguishable from a thyroglossal duct cyst. Closely preoperative evaluations are advised for patients who are discovered to have a thyroglossal duct cyst, including a complete physical examination, accurate head and neck examination, palpation of the thyroid gland, thyroid function tests, a thyroid scan and CT/MRI scan. Malignancy should be suspected if the cyst becomes hard, fixed, irregular, or suddenly expanding with palpable neck lymph nodes. CT/MRI scan, meanwhile, may show a solid mass with invasive features, such as local invasion, irregular border, and calcification of cyst [6]. Also, thyroglossal duct carcinomas should also be distinguished from carcinoma arising from the tip of the pyramidal lobe [7]. In the present case, preoperative evaluation, as well as pathological examination of the thyroid, showed no pathologic findings in the thyroid gland.

The use of FNAC has currently become a common preoperative diagnosis for tumors. By far, just few cases of thyroglossal duct cyst carcinoma was diagnosed preoperatively [8,9]. FNAC, particularly, if performed under ultrasound guidance, may improve the diagnostic value of this exam [10].

The definitive management of thyroglossal duct cyst carcinoma remains controversial, with concern regarding the treatment strategy for the management of thyroid gland and neck dissection. The common surgical procedure used for thyroglossal duct cysts is Sistrunk's procedure, involving removal of the entire cyst and the central portion of the hyoid bone, and a core of tissue around

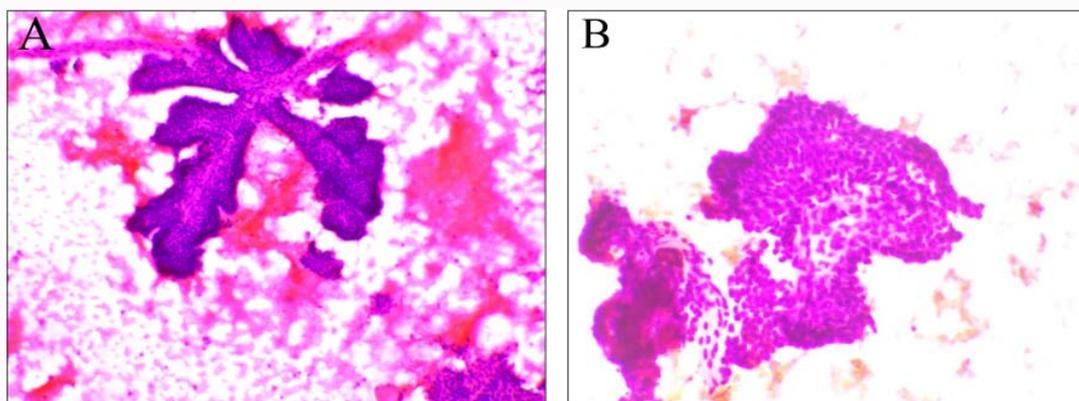


Figure 2: Fine-Needle Aspiration (FNA) and cytology of the solid-cystic lesion and lymph nodes. (A) The tumor was clarified as papillary carcinoma; (B) metastasizing papillary carcinoma was found in the bilateral swelling lymph nodes. (HE x400).

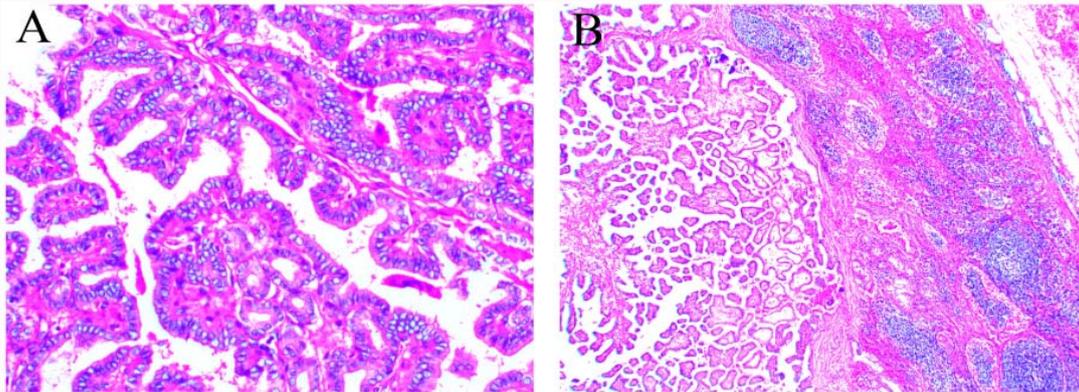


Figure 3: The histopathology of the surgical excision specimen. (A) Specimens confirmed the diagnosis as an invasive primary thyroglossal papillary carcinoma. (HE x400) (B) Metastasis signal was found in cervical lymph nodes. (HE x200).

the thyroglossal tract to open into the oral cavity at the foramen cecum [11,12]. The classic Sistrunk's operation suffices for non-metastatic disease and medically low-risk patients, while high-risk patients that identify with intrathyroidal metastasis, local invasion or nodal metastasis, are recommended to undergo an additional total thyroidectomy with neck dissection and postoperative radioactive iodine ablation therapy and suppressive thyroxine therapy [13].

In this case, the patient received total thyroidectomy and Sistrunk's procedure, and last but not least, because of the evidence of cervical lymph node involvement, the patient performed radical neck dissection, as well as complementary iodine therapy.

The prognosis of papillary carcinoma arising in a thyroglossal duct cyst carcinoma is excellent, with an overall survival rate of 95.6% at 10 years, and the frequency of regional nodal metastasis and local invasion is much lower in papillary carcinomas arising in the thyroglossal duct cyst [1,8]. Careful long-term follow-up, consisting of physical examination, ultrasound of the surgical region and thyroid, and total body scintigraphy, is important for papillary carcinoma. It recommended that all patients must have a neck scan and be reassessed every six months during the first year and annually after operation [14].

Conclusion

What we reported here adds to the published literature of cases of thyroglossal duct cyst carcinoma. Preoperative diagnosis helps determine the operation performance for those patients with midline tumors. For this reason, in such patients, computerized tomography, thyroid scintigraphy and thin-needle aspiration biopsy are referred to be done if necessary, and the proper treatment protocols should carry out after a definite diagnosis.

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