



ABSTRACT

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PAPILLARY CARCINOMA OF THE THYROGLOSSAL DUCT CYST: A CASE REPORT

Introduction. Thyroglossal duct cysts (TGDCs) can be diagnosed at any age. They are most common in women aged 30–40. Cysts can become malignant in 1% of patients. As a result, a very rare tumor – TGDC carcinoma – develops. Some oncologists believe it is sufficient to perform surgery using the Sistrunk procedure. Other scientists insist on performing the Sistrunk procedure followed by thyroidectomy, lymph node dissection, and radioactive iodine ablation. In this report, we would like to share our successful experience in treating a patient with papillary carcinoma of PCOS.

Case report. A 47-year-old woman had persistent midline neck swelling for three years. This lesion was completely asymptomatic. The patient had no cases of head and neck irradiation or a family history of thyroid gland diseases. Physical examination revealed a solid, movable, smooth mass measuring 6.0 × 5.0 × 4.0 cm on the anterior surface of the neck. The thyroid gland was of normal size, without nodules. Lymph nodes of the neck were not enlargement. During sticking out the tongue and swallowing, the tumor moved upwards. Neck ultrasound, CT scan, and fine-needle aspiration biopsy confirmed the presence of TGDC papillary carcinoma. All laboratory results, including thyroid hormones, were within normal ranges.

The patient underwent the Sistrunk procedure, total thyroidectomy with central lymph node dissection, radioactive iodine ablation, and suppression of thyroid function with levothyroxine. The patient's follow-up period has been currently 110 months. There were no signs of relapse.

Discussion. The etiology of the malignant transformation of the TGDC remains unknown. One of the main theories is the spontaneous development of ectopic thyroid tissue located in the cyst walls. Ultrasound, CT, and MRI are considered sufficiently informative methods. A fine-needle aspiration biopsy confirms TGDC carcinoma in 53% of patients before surgery. Virtually all patients with TGDC carcinomas undergo the Sistrunk procedure. In addition, some of them perform a total thyroidectomy. Criteria for total thyroidectomy are age older than 45 years, history of neck irradiation, tumor size greater than 4 cm, presence of regional metastases, and thyroid gland dysfunction.

Postoperative radioiodine ablation and suppressive levothyroxine therapy are considered the best treatment before the start of the follow-up period.

Conclusions. The treatment of patients with TGDC carcinomas should be personalized. Determining criteria should be the patient's age, gender, condition of the thyroid gland and regional lymph nodes, size of the primary tumor, resection margins, invasion of the tumor into the surrounding tissues, and family history of thyroid diseases.

Keywords: thyroglossal duct, cyst, papillary carcinoma, Systrank procedure, thyroidectomy.

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ПАПІЛЯРНА КАРЦИНОМА КІСТИ ЩИТОПОДІБНО-ЯЗИКОВОЇ ПРОТОКИ (ВИПАДОК З ПРАКТИКИ)

Вступ. Кісти щитоподібно-язикової протоки (КЩЯП) можуть діагностуватися у будь-якому віці. У 1% пацієнтів кісти можуть малігнізуватися. Як наслідок, розвивається дуже рідкісна пухлина – карцинома КЩЯП. Частина онкологів дотримується думки, що для радикального лікування достатнім є виконання хірургічної операції методом Сістранка. Інші науковці наполягають на виконанні процедури Сістранка з наступною тиреоїдектомією, шийною лімфатичною дисекцією та абляцією радіоактивним йодом. У цьому звіті ми хотіли б поділитися власним успішним досвідом лікування пацієнта з папілярною карциномою КЩЯП.

Випадок з практики. У 47-річної жінки три роки спостерігався постійний набряк шиї по середній лінії. Це ураження протікало абсолютно безсимптомно. Випадків опромінення голови та шиї та захворювань щитоподібної залози в сімейному анамнезі пацієнта не було. Під час фізикального обстеження виявлено тверде рухливе гладке утворення розміром 6,0 см × 5,0 см × 4,0 см на передній поверхні шиї. Щитоподібна залоза була нормальних розмірів, без вузлів. Лімфатичні вузли шиї не пальпувалися. Під час висовування язика і ковтання пухлина зміщувалася вгору. Ультразвукове дослідження шиї, КТ та тонкогілкова аспіраційна біопсія підтвердили наявність папілярної карциноми КЩЯП. Всі результати лабораторного дослідження, включаючи тиреоїдні гормони, були в нормі.

Пацієнтці виконано операцію Сістранка, тотальну тиреоїдектомію з центральною лімфодисекцією, абляцією радіоактивним йодом та пригнічення функції щитоподібної залози левотироксином. Період спостереження за пацієнткою на даний час становить 110 місяців. Ознак рецидиву не спостерігалось.

Обговорення. Етіологія злоякісної трансформації КЩЯП залишається невідомою. Одна з основних теорій – це спонтанний розвиток ектопічної тканини щитовидної залози, що розташована в стінках кіст. УЗД, комп'ютерна та магнітно-резонансна томографії вважаються достатньо інформативними методами дослідження. Тонкогілкова аспіраційна біопсія дозволяє на

доопераційному етапі підтвердити карциному КЩЯП у 53 % пацієнтів. Практично усі пацієнти з карциномами КЩЯП підлягають процедурі Сістранка. Крім цього, частині з них виконують тотальну тиреоїдектомію. Критерієм для тотальної тиреоїдектомії є вік старше 45 років, опромінення ділянки шиї в анамнезі, розмір пухлини більше 4 см, наявність регіонарних метастазів, дисфункція щитоподібної залози. Післяопераційна абляція радіоактивним йодом та супресивна терапія левотироксином вважаються найкращим лікуванням перед початком періоду спостереження.

Висновки. Лікування пацієнтів з карциномами КЩЯП повинно бути персоналізованим. Визначальними критеріями повинні бути вік пацієнта, стать, стан щитоподібної залози та регіонарних лімфатичних вузлів, розмір первинної пухлини, краї резекції, інвазія пухлини в оточуючі тканини та анамнестичні дані щодо захворювань щитоподібної залози.

Ключові слова: щитоподібно-язикова протока, кіста, папілярна карцинома, процедура Сістранка, тиреоїдектомія.

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How to cite / Як цитувати статтю: Duzhiy I, Kononenko M, Moskalenko Yu, Zhukova K. Papillary carcinoma of the thyroglossal duct cyst: a case report. *East Ukr Med J.* 2023;11(2):121-126

DOI: [https://doi.org/10.21272/eumj.2023;11\(2\):121-126](https://doi.org/10.21272/eumj.2023;11(2):121-126)

INTRODUCTION / ВСТУП

Thyroglossal duct cysts (TGDCs) can be diagnosed at any age. They are most common in women aged 30–40. Cysts can become malignant in 1% of patients. Therefore, the development very rare type of tumor – TGDC carcinoma. The most common histological variant is papillary cancer (92%) [1].

The thyroid gland begins to develop in the third week of embryogenesis. The thyroglossal duct (TGD) attaches the diverticulum of the thyroid gland to the tongue. After the fifth week of gestation, its involution begins. In the eighth week, the TGD disappears. Nevertheless, in 7% of patients, it remains as a cyst, duct, or ectopic tissue. Genetic factors are believed to be the leading cause of TGDC [2].

Although the malignant transformation of cysts of TGDC rarely occurs, many case reports are described in the scientific literature [3, 4]. However, there still needs to be a clear approach to treating patients. Some oncologists believe it is sufficient to perform surgery using the Sistrunk procedure. Other scientists insist on performing the Sistrunk procedure followed by thyroidectomy, lymph node dissection, and radioactive iodine ablation. In this report, we would like to share our successful experience treating a TGDC papillary carcinoma.

Case report. A 47-year-old woman had three years of persistent midline neck swelling. This lesion was completely asymptomatic. The patient had no cases of head and neck irradiation or a family history of thyroid gland diseases. The patient provided written informed consent for the publication of this clinical case report.

Physical examination revealed a solid, movable, smooth mass measuring 6.0 × 5.0 × 4.0 cm on the anterior surface of the neck. The thyroid gland was of normal size, without nodules. Lymph nodes of the neck were not enlargement. During sticking out the tongue and swallowing, the tumor moved upwards.

Ultrasound examination of the neck revealed a 5.4 × 4.2 × 3.0 cm heterogeneous mass with thickened walls located subcutaneously just below the hyoid bone. No signs of lymphadenopathy were found. A fine-needle aspiration biopsy of the anterior neck cyst under ultrasound control confirmed the presence of papillary carcinoma of the TGDC.

Computed tomography of the neck revealed a heterogeneous irregular lesion at the hyoid bone and thyroid cartilage level with calcification. All laboratory results, including thyroid hormones, were in normal ranges.

The patient underwent a Sistrunk procedure (complete resection of the TGDC carcinoma and the

body of the hyoid bone). Considering the patient's age and the tumor's size, total thyroidectomy with central lymph node dissection was performed. The pathomorphological examination confirmed the diagnosis of papillary carcinoma of the TGDC with metastases in the lymph nodes of the central neck compartment (level VI) (Fig. 1). Histological

examination of the thyroid gland revealed autoimmune thyroiditis without atypia. The margins after surgery were negative. No signs of lymphovascular or perineural invasion were found. The tumor did not invade the adjacent fat tissue, muscles, or hyoid bone.

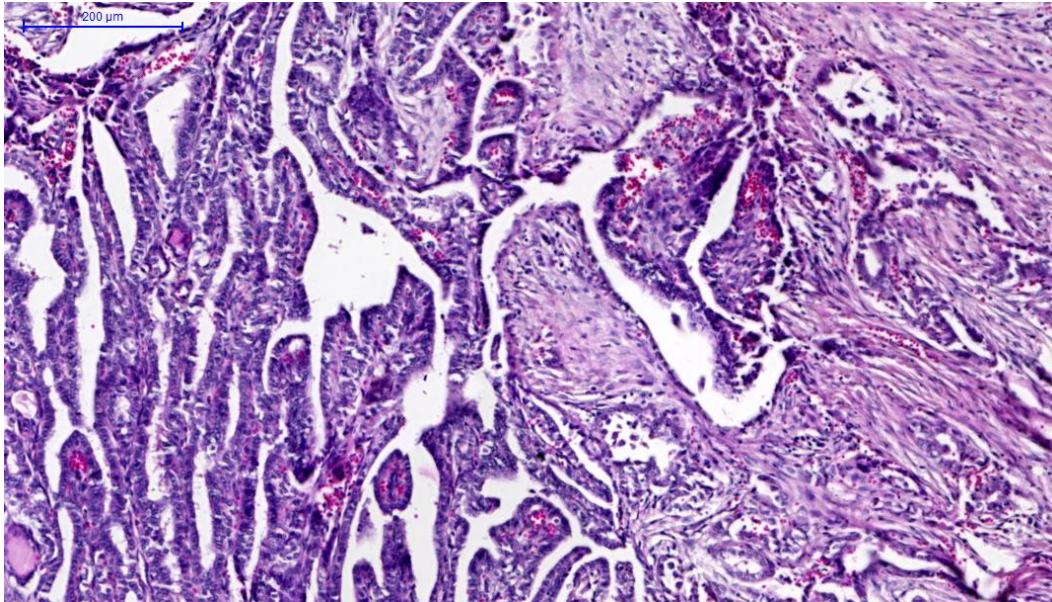


Figure 1 – Pathohistological examination of the TGDC showed a papillary thyroid carcinoma (degree of magnification is 100x, hematoxylin and eosin staining)

Ablation with radioactive iodine was started one month after surgery. Thyroid suppression with levothyroxine was performed after I131 ablation. The patient's follow-up period is currently 110 months. There were no signs of disease relapse.

Discussion. The etiology of the malignant transformation of the TGDC remains unknown. One of the main theories is the spontaneous development of ectopic tissue of the thyroid gland, which is in the walls of cysts. The absence of medullary carcinomas of TGDC confirms this conclusion [1].

Our patient had the most common histological variant of TGDC carcinomas – papillary type. However, some patients can have mixed (8%) or squamous cell carcinoma (6%). Follicular and anaplastic carcinomas develop even more rarely [2].

7–15% of patients have metastases in the lymph nodes of the neck. The frequency of regional metastases is lower than in patients with papillary thyroid cancer. Recurrence of the disease is registered in 4.3% of people. However, the mortality rate in patients with TGDC carcinoma is extremely low [1].

Ultrasound, CT, and MRI are sufficiently informative methods. The heterogeneity of the cyst and the presence of a solid component indicate TGDC carcinoma. Thickening of the cyst walls, irregular border, and biomineralization also confirm the diagnosis of cancer [5]. The ultrasound and CT features of our patient's TGDC papillary carcinoma were a heterogeneous mass with thickened walls, irregular borders, and biomineralization.

A fine-needle aspiration biopsy allows for confirmation of TGDC carcinoma in 53% of patients before surgery. In the case of a solid component, the diagnostic value of this method increases [5].

According to the meta-analysis data, 98.9% of patients with TGDC carcinomas underwent the Sistrunk procedure. Almost all scientists approve of this approach to surgical treatment. In 61% of patients, the Sistrunk procedure was followed by total thyroidectomy [1].

Proponents of the theory of performing only the Sistrunk procedure insist that total thyroidectomy does not affect the outcome of treating TGDC carcinomas. It increases the risk of damage to the

laryngeal nerve and hypocalcemia. Moreover, in univariate regression analysis, only the Sistrunk procedure was a predictor of overall survival [6]. Rayess et al. [1] declare that total thyroidectomy does not need to be performed if the patient meets the following criteria: age younger than 45 years, no history of neck irradiation, tumor size less than 4 cm, no regional metastases, and normal thyroid function.

Our patient was over 45 years old, and the tumor size was 5.4 cm. In addition, according to the pathohistological examination, metastases were

found in the lymph nodes of the VI level of the neck. According to the criteria of Lancini and others [7], such a patient has a high risk of recurrence, so she needs to perform the Sistrunk procedure, total thyroidectomy, and ablation with radioactive iodine. In addition to the specified criteria, indications for this treatment are male gender, distant metastases, and tumor invasion into the surrounding tissues. Postoperative ablation with radioactive iodine and suppressive therapy with levothyroxine is considered the best treatment before starting follow-up [8].

CONCLUSIONS / ВИСНОВКИ

TGDC carcinomas are very rare tumors. Treatment of patients should be personalized. Determining criteria should be the patient's age,

gender, condition of the thyroid gland and regional lymph nodes, size of the primary tumor, resection margins, invasion of the tumor into the surrounding tissues, and family history of thyroid diseases.

CONFLICT OF INTEREST / КОНФЛІКТ ІНТЕРЕСІВ

The authors declare no conflict of interest.

FUNDING / ДЖЕРЕЛА ФІНАНСУВАННЯ

None.

AUTHOR CONTRIBUTIONS / ВКЛАД АВТОРІВ

All authors substantively contributed to the drafting of the initial and revised versions of this paper. They take full responsibility for the integrity of all aspects of the work.

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Received 24.04.2023

Accepted 07.06.2023

Одержано 24.04.2023

Затверджено до друку 07.06.2023

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