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Ectopic thyroid gland microcarcinoma with metastasis to the cervical lymph nodes and lungs. Case report

Summary

The carcinoma of the thyroglossal dust is rare tumor. The 15-year old female patient underwent Sistrunk procedure with diagnosis of the cyst in the midline of neck. The pathological examination revealed thyroid papillary microcarcinoma. Patient were referred to National Oncology Center for further evaluation. After the ultrasonography, the pathologic lymph node in the lateral right side of the neck was detected and biopsied with the results of suspicious for malignancy. The thyroidectomy with modified neck dissection on the left side was performed. After operation on the scintigraphy and SPECT-CT there was multiple metastasis detected in both lungs.

The ectopic thyroid, which is rare in clinical practice, is the result of defect in different stages of thyroid embryogenesis. The clinical symptoms depend on localization, dimensions, functions and morphology. The risk of malignization is 1%. The treatment in ectopic carcinoma is the same like in normal thyroid carcinoma.

Key words: thyroglossal duct cyst; thyroid papillary microcarcinoma; ectopic thyroid cancer.

During embryogenesis, on the 3th and 4th week, the thyroid developed in the posterior part of tongue, from the endodermal diverticulum of the middle part of primitive pharynx and after this migrate by the thyroglossal duct (base of tongue-foramen cecum, sublingual bone, larynx, trachea) toward anterior part of larynx to the pretracheal area and reaches its anatomic location on 7th week. The thyroglossal duct after 7th week begins to atrophy.

The ectopic thyroid is the result of diverticulum migration defect of the tongue base and absent of atrophy of the thyroglossal duct. This occur in 1 in 100 000 - 300 000 of population. First time this was described by Hickman in 1869, when newborn died 16 hours after delivery from asphyxia since his thyroid was located in the base of the tongue and compressed epiglottis [1, 20]. Incomplete atrophy of the thyroglossal duct may result in residual ectopic thyroid tissue or formation of the cyst. The thyroglossal cyst described in 1911 by Brentano and in 1915 by Ucherman like asymptomatic lesion. Now these kind of ectopic thyroid abnormalities are rare. Ectopic thyroid carcinoma is very rare [2, 3, 27].

Ectopic thyroid tissue is located mostly in the lingual (90%), sublingual, thyroglossal, laterocervical, rare submandibular, retroperitonal, mediastinal region and in the pancreas [2, 18, 37]. In 70-75% cases when ectopic lingual thyroid takes place the thyroid is absent in its regular location. The ectopic thyroid has the same function and also can have inflammation, hyperplasia and malignization [18, 32].

The thyroglossal cyst is the most frequent ectopic pathology and it is 70% of the time localized in the midline of the neck in children and 7% in adults. The risk of malignization is 1% [4, 6, 11, 14, 17]. To prevent residue of thyroglossal cyst Sistrunk procedure (resection of the part of hyoid bone) should be performed [11, 33, 41].

Most cases of thyroglossal duct carcinoma (TGDC) were diagnosed in postoperational pathological specimens of patients, operated due to thyroglossal duct cyst [5, 15, 42].

The cure rate for papillary thyroid carcinoma in TGDC was reported as 95% when treated by Sistrunk procedure (SP). Furthermore, in a recent publication, the survival rate was reported as 100 % when treated by SP/ thyroidectomy with lymph node dissection. The surgical extent and the use of post-operative radioactive iodine (RAI) therapy remain controversial. Recently, the concept of stratification by risk was proposed to identify patients who would benefit from more aggressive treatment, *i.e.* total thyroidectomy and/or neck dissection and RAI therapy [16, 45,46].

We want to share cases of ectopic thyroid carcinoma of patients in National Center of Oncology.

Fist clinical case: The patient, 15 year-old female, was admitted to the private clinic because of lesion in the midline of neck. The diagnosis of «midline neck cyst» was established and the patient underwent Sistrunk procedure. After pathological investigation of the surgical specimen, there was papillary carcinoma detected. The tumor diameter was 0.8 cm, it was small foci of calcinations found; no any necrosis, high mitotic activity or hemorrhage detected. After diagnosis, the patient was referred to the National Center of Oncology. On the ultrasound there were no pathological lesions found in the normal projection of the thyroid gland. On the right side of the neck there was slightly changed lymph node measured 1.5 cm. The fine needle aspiration was performed and the malignant suspicious cells were detected. Then, thyroidectomy and right side modified neck dissection was performed. On the pathological examination there were diffuse hyperplasia of thyroid gland, metastasis of papillary carcinoma in one lymph node and reactive hyperplasia in other lymph nodes reported (Figure 1 a, b).

4 weeks after the operation (TSH-100mU/l, Tg-16ng/l) 125mCi radioactive iodine therapy began. 1 week after radioactive iodine therapy scintigraphy of the whole body was done and with the

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minimally residual tissue in the operation site, also multiple focal depositions in both lungs were revealed. To establish the nature of these depositions, CT and SPECT-CT (single photon emission computed tomography and computed tomography) of the lungs were performed. While on the CT there were no pathology in the lungs detected, on the SPECT – CT there were multiple metastases in the both lungs found (Figure 2 and Figure 3 a, b). The TSH suppressive treatment was prescribed to the patient.

The second clinical case: 57 year-old female, suffering from the large neck tumor, was admitted to clinic with complaints on breathlessness and from time to time asphyxia. The history of the lesion was reported for 10 years. On the Ultrasound and CT the lesion was detected in the anterior submandibular region, intimal adjoining to the anterior muscles of larynx, with calcinations and cystic-necrotic components measured 53x37 mm (Figure 4 a, b). The clinical diagnosis was thyroglossal cyst. The dimensions of the thyroid were normal, parenchyma – heterogenic. There were no enlarged lymph nodes found.

The fine needle aspiration was performed and the cytological examination revealed suspicion for malignant cells.

The Sistrunk procedure was performed for this patient where the pathological examination revealed papillary carcinoma 5 cm in diameter. Later, it was decided to perform thyroidectomy and the diagnosis was Hürtle cell adenoma with reactive hyperplasia in 2 lymph nodes (igure 5 a, b, c).

In the postoperative period TSH -100mIU/l, Anti-Tg – 52.4 bV/ml, Tg – 0.3 nq/ml, the patient received 150 mCi radioactive iodine treatment and after it was completed the TSH suppressive therapy was prescribed.

Discussion: The thyroglossal carcinoma is rare malignant but when it is malignant then papillary (75-85%), follicular variant papillary carcinoma (7%), very rare anaplastic carcinoma and tended to have the worst prognosis. In the literature there is no medullary carcinoma found, which can be explained by the embryologic derivation [6, 10, 16, 18, 34, 47].

The papillary microcarcinoma metastasis was found in 10-20% [35, 36]. In literature, depending of metastasis, unilateral or bilateral lymphodissection was performed [7, 15, 22, 26, 33]. Distant metastasis occurred in 1.3% of cases, mostly to lungs, bones, brain and mediastinum [13, 15, 16, 27]. The ectopic thyroid carcinoma very rare metastasize to bilateral cervical lymph nodes or submental region. Cervical lymph node metastasis from papillary carcinoma of a TGDC was reported in 7-25% of cases, and distant metastasis was described in 1.3% of cases. In the cases where it metastasized then the bilateral neck dissection with iodine therapy and future TSH suppression are recommended. [15, 16, 21, 22, 30, 43, 47]. Including this fact, during diagnostic of the cervical tumors, the ectopic thyroid carcinoma should be taken into account [7, 8, 9, 19].

Scintigraphy, using Tc-99m, I-131, or I-123, is the most important diagnostic tool to detect ectopic thyroid tissue and shows the absence or presence of thyroid in its normal location.

Thyroid scan can also unmask additional sites of thyroid tissue. It is both sensitive and specific for differentiation of an ectopic thyroid from other causes of midline neck masses [20].

That radionuclide thyroid scanning and function testing may be useful not only for the diagnosis of an ectopic thyroid but also before deciding on the therapeutic modality; patients should be followed up to detect changes in thyroid function and malignant transformation [32].

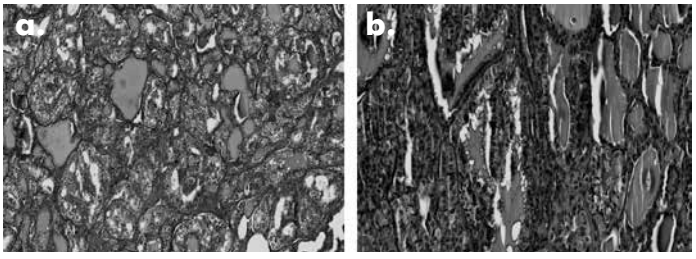

Figure 1.

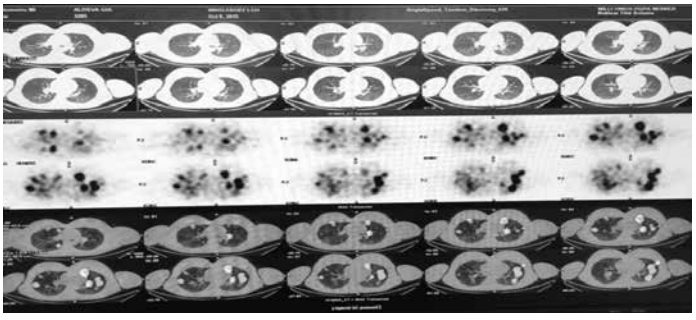

Figure 2. Comparative pictures of lungs CT, scintigraphy SPECT-CT.

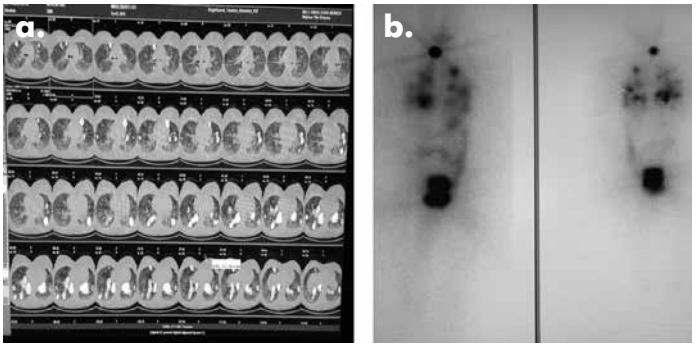

Figure 3. a) SPECT-CT examination: multiple metastasis in both lungs b) whole body scintigraphy

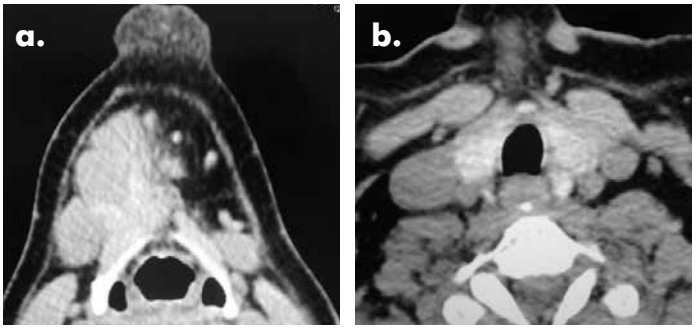

Figure 4.

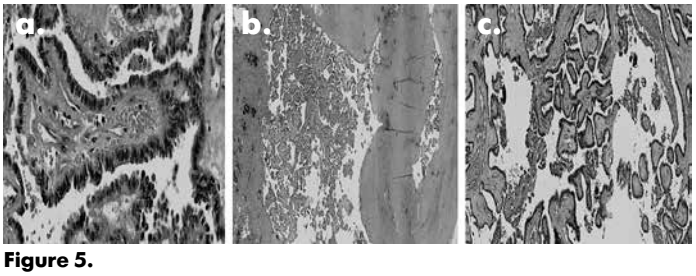

Figure 5.

CT and MRI are valuable tools in identifying the site of ectopy, especially when it is distant from the descending pathway of thyroid. In cases of cervical lymphadenopathy or intratracheal invasion by the tumor, CT and MRI may assist as the suspicion for increased malignancy [2, 20, 40, 44].

The last research works shown, that preoperative diagnosis of thyroglossal cyst and thyroglossal carcinoma is easier in adults than in children. The calcifications of the cyst wall found on CT, MRT are suspicious for malignant transformation. The cyst wall invasion was found in 21% of patients [22, 39]. In the adults, the CT, MRT and FNA (fine needle aspiration) give possibility to establish diagnosis and the treatment can be planned [12, 15, 16, 31]. We recommend the fine needle biopsy of the possible thyroglossal cyst and the lymph nodes if present. [15, 29]. Some researchers showed, that solid component in the cyst found in preoperative Ultrasound can be one of the carcinoma signs and should be monitored carefully. [28].

Fine needle aspiration cytology (FNAC) provides considerable assistance in confirming the diagnosis of ectopic thyroid. It is the only modality to differentiate between a benign and a malignant lesion. However, FNAC results may sometimes be misleading or non-diagnostic, especially in cystic masses [8, 20].

Some authors advocate (Bosch və Livolsi) that if the thyroglossal carcinoma is inside of the cyst without invasion then Sistrunk procedure is enough, but if the tumor invades the cyst capsule, in this cases thyroidectomy is required [23, 24]. Other authors advocate on thyroidectomy additional to thyroglossal cyst surgery since during carcinoma diagnosis the TSH levels should be suppressed and be between 0.1-0.5 mIU/L. Also, after thorough evaluation of the bilateral neck with the help of ultrasound, MRI and fine needle biopsy when necessary, the patient might require modified neck dissection [15, 25, 38].

O'Connell et al. suggest the use of USG and CT scan or MRI with FNA and justify the use of total thyroidectomy and or lymphnode dissection in case significant doubt remains. They also suggest for additional therapy in the form of ^{131}I ablation in case of thyroid gland involvement, soft tissue extension and lymph node involvement as it is standard practice for the management of well differentiated thyroid carcinoma [25].

Regional lymph node metastasis was reported in 7.7–12.9 % of the malignant cases. Since the case reports with neck metastases in case of TGDC (thyroglossal duct cyst) papillary is limited, we do not have standard treatment protocol. Literature is deficient in whether we should choose unilateral or bilateral neck dissection and also if one should perform modified neck dissection or selective nodal excision. Yamada et al. reported a case of TGDC papillary carcinoma with bilateral neck metastases. They performed en-bloc resection of submental tumor and concurrent bilateral supraomohyoid neck dissection avoiding total thyroidectomy [7, 15, 21, 22, 26, 33, 43].

A slightly different approach is recommended by Patel et al. [16], who examined specific patient characteristics and sought to stratify them into «low risk» and «high risk.» A patient younger than 45 with a tumor less than 4 cm in size without soft tissue extension and without distant metastases and a clinically and radiologically normal thyroid gland is considered «low risk» and can be treated with the Sistrunk procedure alone (16, 48). Those in a higher risk group (older than 45, tumor larger than 4 cm, with soft tissue extension, with nodal or distant metastases) require more aggressive treatment, including Sistrunk procedure, total thyroidectomy with or without neck dissection, followed by radioactive iodine therapy. However, the small sample size may make any risk group stratification inaccurate. We recommend that specific consideration be given to each individual patient.

The ectopic thyroid, which is rare in clinical practice, is the result of defect in different stages of thyroid embryogenesis. The clinical symptoms depend on localization, dimensions, functions and morphology. The risk of malignisation is 1%. The treatment in ectopic carcinoma is the same like in normal thyroid carcinoma.

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Резюме

Эктопическая микрокарцинома щитовидной железы с метастазами в шейные лимфатические узлы и легкие. Клинический случай

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Карцинома тиреоглосального протока – новообразование, которое редко встречается в клинической практике. В приведенном клиническом случае речь идет о больной в возрасте 15 лет, которой по поводу кисты шейного отдела была проведена операция Систрунка. В результате проведенного патоморфологического обследования была обнаружена микрокарцинома щитовидной железы. Пациентка была направлена в Национальный онкологический центр, где с помощью УЗ-исследования были обнаружены патологически измененные лимфатические узлы. Больной было проведено оперативное вмешательство – тиреоидэктомия, селективная шейная лимфодисекция. После операции при проведении сцинтиграфии и ОФЭКТ СТ в обоих легких были обнаружены множественные метастазы.

Редко встречаемое эктопическое образование щитовидной железы является результатом дефекта на разных стадиях эмбриогенеза щитовидной железы. Клинические симптомы зависят от локализации, размера, функции и морфологии эктопического тиреоида. Риск малигнизации – 1 %. В случае эктопической карциномы щитовидной железы применяется та же тактика лечения, что и при обычной карциноме щитовидной железы.

Ключевые слова: щитовидно-языковой проток, киста, папиллярная микрокарцинома щитовидной железы, эктопический рак щитовидной железы.

Резюме

Ектопічна мікрокарцинома щитоподібної залози з метастазами в шийні лімфатичні вузли і легені. Клінічний випадок

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Карцинома тиреоглосальної протоки – новоутворення, яке рідко зустрічається у клінічній практиці. В наведеному клінічному випадку йдеться про хвору у віці 15 років, якій з приводу кісти шийного відділу була проведена операція Систрунка. У результаті проведеного патоморфологічного обстеження була виявлена мікрокарцинома щитоподібної залози. Пацієнтка була направлена до Національного онкологічного центру, де за допомогою УЗ-дослідження були виявлені патологічно змінені лімфатичні вузли. Хворій було проведено оперативне втручання – тиреоїдектомія, селективна шийна лімфодисекція. Після операції при проведенні сцинтиграфії і ОФЕКТ в обох легенях були виявлені множинні метастази.

Ектопічне утворення щитоподібної залози, що рідко зустрічається, є результатом дефекту на різних стадіях ембріогенезу щитоподібної залози. Клінічні симптоми залежать від локалізації, розміру, функції і морфології ектопічного тиреоїду. Ризик малігнізації – 1 %. У випадку ектопічної карциноми щитоподібної залози застосовується та сама тактика лікування, що й при звичайній карциномі щитоподібної залози.

Ключові слова: щитоподібно-язикова протока, кіста, папілярна мікрокарцинома щитоподібної залози, ектопічний рак щитоподібної залози

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