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A Young Girl with Ectopic Thyroid Papillary Carcinoma

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Abstract

Ectopic thyroid tissue is a form of thyroid dysgenesis, mainly found in the base of the tongue. Ectopic thyroid tissue is generally benign and rarely occurs with a eutopic thyroid gland. We introduce the case of a 16-year-old girl diagnosed with papillary thyroid carcinoma, found in a thyroglossal duct cyst. This case was suspected at first of malignancy due to the presence of echogenicity within the cyst. After the surgical removal of the thyroglossal duct cyst by the Sistrunk procedure, the histological diagnosis was papillary thyroid carcinoma. Due to the tumor's size of 3 cm, the Sistrunk procedure was followed by total thyroidectomy and radioactive iodine therapy. The histological evaluation of the resected thyroid gland did not show any malignant cells.

Abbreviations

TGDC: Thyroglossal Duct Cyst; TGDCCa: Thyroglossal Duct Cyst Carcinoma; MRI: Magnetic Resonance Imaging; FNA: Fine Needle Aspiration

Introduction

Ectopic thyroid is defined as any thyroid tissue not located in its normal position. The prevalence of thyroid ectopia is about 1 per 100.000 – 300.000 individuals [1–3]. The most frequent location is the base of the tongue. Other sites include the anterior tongue, submandibular region, sublingual region, larynx, trachea, mediastinum, and heart [2]. Dyspnea and dysphagia are its most common symptoms [3–6]. In children, Thyroglossal Duct Cysts (TGDCs) are the most common congenital anomalies of the thyroid gland. They present as a painless mass in the anterior part of the neck. The majority of them are diagnosed during the clinical examination [7–9]. In any such case, ultrasound and/or Magnetic Resonance Imaging (MRI) of the neck are the recommended examinations [9–11]. In the majority of cases, the histological examination highlights a thyroid carcinoma after the surgical removal of a thyroglossal duct cyst by the Sistrunk procedure [6,10,11]. There is no common therapeutic guideline yet regarding the possible thyroidectomy and/or additional radioiodine therapy for these patients. Every case needs an individual approach depending on the patient's age, the tumor's size, the histopathological type of cancer, the distance of the resection border to the malignant cells, and the morphological and functional condition of the thyroid gland [9,11,12].

Case Presentation

A 16-year-old girl presented at our clinic for a 3-month history of a painless soft mass located in the anterior part of her neck. She had only shortness of breath. The physical examination found a

mobile nodule of approximately 3 cm.

During the neck ultrasonography (Figure 1), in the cervical anterior median line region, in the middle 1/3, a cystic lesion up to 30 mm x 25 mm was evident. Irregular echogenic vegetation was seen in it, along with vascularization after color Doppler to determine the following: complex cyst of the thyroglossal duct was suspected. The structure of the thyroid gland was isoechoic, without nodules, with normal vascularization. Regional lymphadenopathy was not evident. Therefore, the mass was considered suspicious for neoplastic tissue. The radiologist recommended an MRI of the head and the neck.

The MRI of the head and the neck (Figure 2) was performed with a 1.5 Tesla head and neck MR scanner, with FSE, STIR, and FAT-SAT sequences (T1, T2). In the medial region of the neck, within the sublingual region, and in the context of the soft tissues: a nodule with dimensions in the axial plane of about 26 mm x 16 mm with a craniocaudal extension of about 20 mm was evident. Along with cystic aspects, but lobulated, and the presence of soft tissue with septa inside it. This nodule appeared to be closely related to the digastric and mylohyoid muscles and the hyoid bone. There were no lesions in the thyroid gland, without evidence of bilaterally focal lesions in the submandibular and parotid glands. Paranasal sinuses with normal pneumatization in the planes were included in the examination. The patient underwent specific thyroid function blood tests, from which the thyroid showed normal function [FT3 – 51.1 (23 pmol/l – 62 pmol/l), FT4 - 13.0 (10.6 pmol/l –19.4 pmol/l), TSH - 1.74 (0.3 µIU/ml –5.0 µIU/ml)] and negative antithyroid antibodies [AntiTPO < 0.8 (< 8.0 IU/ml), AntiTg < 6.4 (< 18 IU/ml)]. Thyroglobulin level was 53.9 (3.5 ng/ml –77 ng/ml). The patient had not previously used any medication for the thyroid gland. One week later, after discussing the case with the ENT surgeon, the Sistrunk procedure was performed.

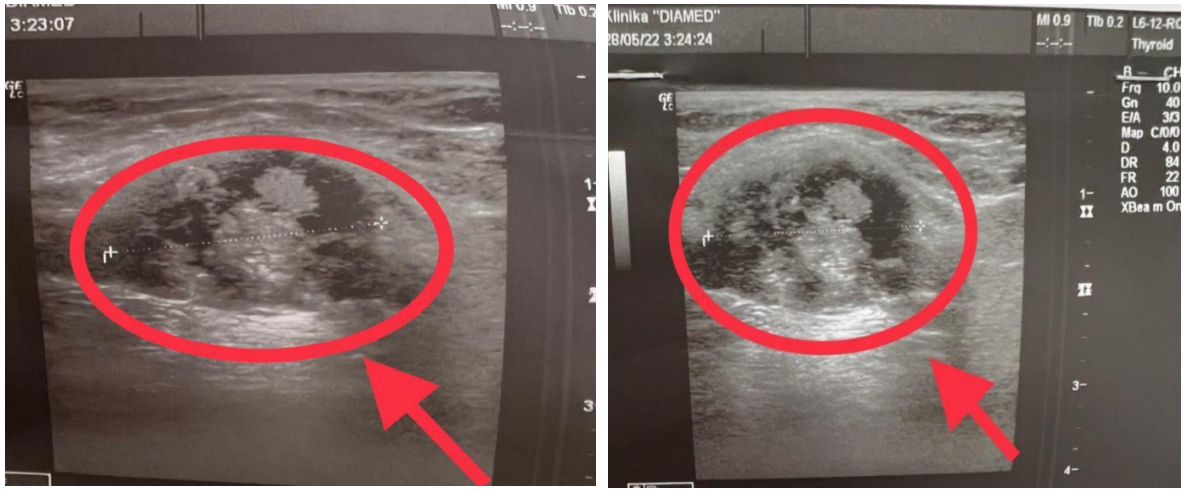

Figure 1: Ultrasound of the neck.

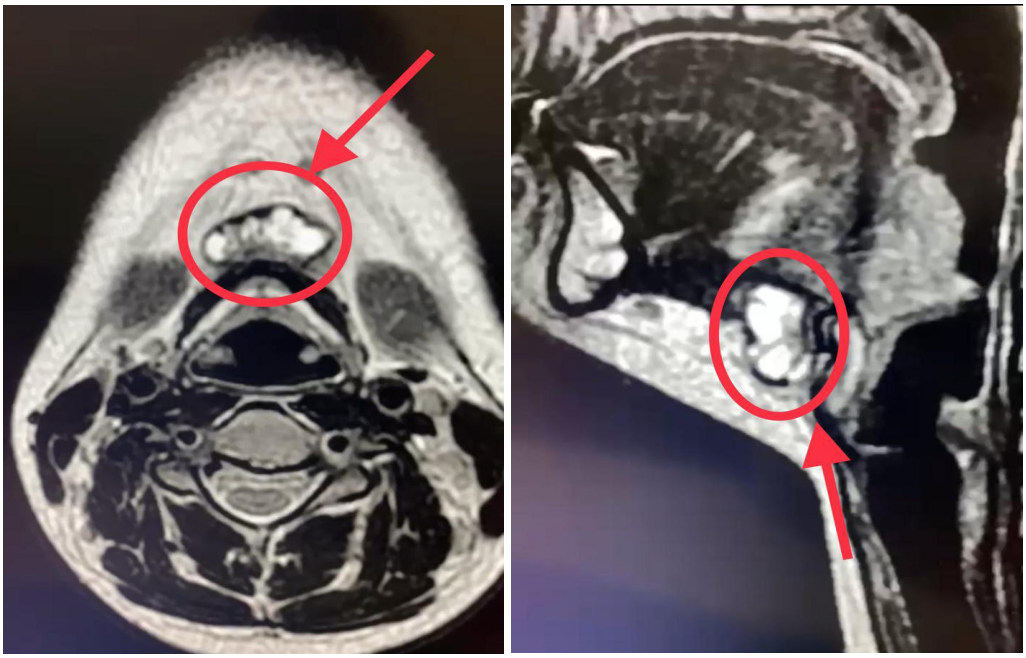

Figure 2: Head and neck MRI.

The histopathological evaluation of the removed thyroglossal cyst noted malignant thyroid cells of a papillary thyroid carcinoma inside it (Figure 3).

In (Figure 3a), we note the mild lymphoplasmacytic infiltration, lymphoid follicle formation, varying sizes of the thyroid follicles with dense colloid, and oxyphilic changes in the follicular epithelium.

Microscopy of the second photo (Figure 3b) displays the epithelium of the cyst. The lining epithelium is a single layer of flattened cells. Around the cyst are thyroid follicles.

In (Figure 3c1), we note the Papillary carcinoma, the true papillae with multiple branching with fibrovascular cores.

In (Figure 3c2), the cells are presenting changes in nuclear size and shaped ground glass nuclei with nuclear membrane irregularity.

Because of these conditions, a total thyroidectomy was performed two weeks later. The resected thyroid gland resulted histologically negative for malignancy. Two weeks after the thyroidectomy, the patient was subjected to radioactive iodine therapy (30 mCi). Currently, the patient is on TSH suppression therapy, with 100 mcg of Levothyroxine per day. The last thyroglobulin level is 0.04 ng/ml. The follow-up plan includes the ultrasonography of the neck and the thyroglobulin level test every six months for a 5-year period.

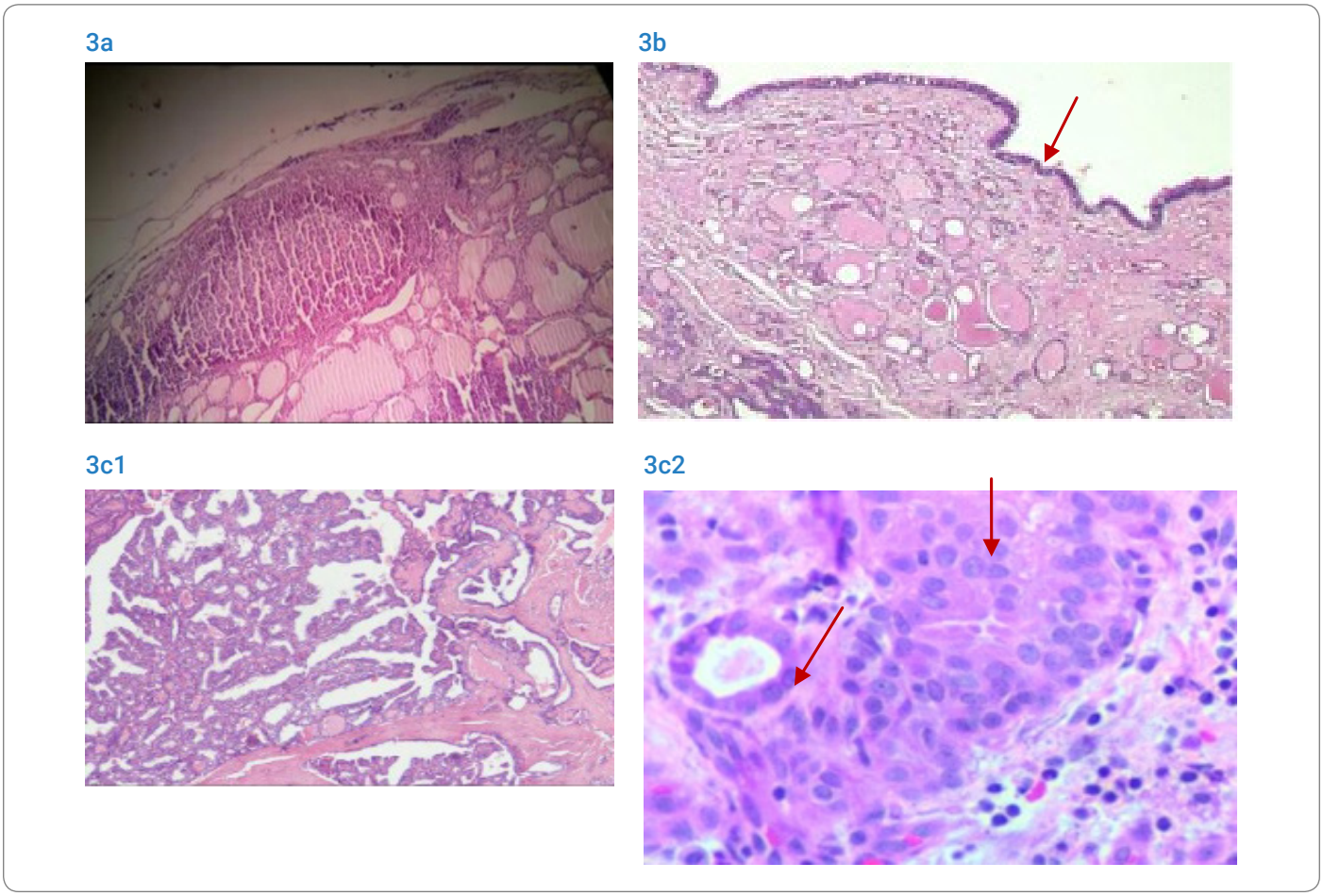

Figure 3: The histopathological description; **a:** H & E, x 40; **b:** H & E, x 40; **c1:** H & E, x 10; **c2**: H & E, x 100.

Discussion and Conclusions

Ectopic thyroid is the most common form of thyroid dysgenesis [1]. The majority of them are found as a component of the thyroglossal duct cysts, commonly asymptomatic and benign [7,9,15,16]. However, every TGDC should be evaluated through a physical examination, ultrasonography and/or MRI of the neck, and thyroid blood tests [9–11]. Papillary thyroid carcinoma is the most common form of TGDCCas, followed by squamous cell carcinomas [9,12–14]. However, their management strategy continues to be indefinite, with controversial opinions about thyroidectomy in cases with normal thyroid morphology on ultrasonography. So, from one side, there are recommendations not to perform a concurrent thyroidectomy but just to remove the tumor of the thyroglossal cyst and to monitor for signs of future thyroid involvement [15,19]. This opinion is supported by Ying-Ying Zhu et al. [20], which recommend the total thyroidectomy and neck dissection only in the presence of thyroid lesions or cervical lymphadenopathy.

On the other hand, Laura Falvo et al. [14], in her study "Papillary thyroid carcinoma in thyroglossal duct cyst: case reports and literature review," recommend associating total thyroidectomy because the carcinoma may be multifocal, and there may be a

lymphatic invasion of the thyroid gland. Besides this, in their opinion, this is the best strategy to ensure a correct follow-up by thyroglobulin measurement. In a similar retrospective study of 26 patients with differentiated thyroid cancer in the thyroglossal duct cysts, Gabriella Pellegriti et al. [21] suggested both thyroglossal duct cyst resection and total thyroidectomy. In principle, from all the literature data we consulted, for a TGDCCa under 1 cm, the Sistrunk procedure is enough. In other cases, the possibility of a total thyroidectomy should be considered, followed by radioactive iodine therapy [7,9,14,17,18,21,22]. Our case presents a TGDCCa of 3 cm in size, suspicious of malignancy in the ultrasonography of the neck. Due to the size of the tumor and the request of the patient's parents for the safest possible therapy, we applied the Sistrunk procedure and thyroidectomy, followed by radioactive iodine therapy. Our decision to perform the total thyroidectomy could have been more correct after applying the Fine-Needle Aspiration (FNA) biopsy, but we could not perform this due to the patient's refusal. Either way, Shahin Areej et al. [22], in a cytopathologic study of 26 cases, point out that TGDCCa has low sensitivity rates of FNA biopsy (56%–62%) compared to primary thyroid cancers (85%), probably due to the aspiration of cystic fluid along with the FNA. Based on all the above, and for cases similar to ours, the total thyroidectomy may be considered a safe long-term therapeutic strategy through easy follow-up care by thyroglobulin measurement and scintigraphy [14,15,18,21,22].

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Consent for publication: The patient and her parents have given their informed written consent for publishing this case report.

Availability of data and material: The authors declare that the data supporting the findings of this study are available within the article.

Conflict of Interest

The authors declare no potential conflicts of interest with respect to the research, authorship, and/or publication of this article. Informed consent was obtained for this publication.

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