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Case Report
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Carcinoma of the Thyroglossal Duct Cyst: A Case Report with Commentary

Mihail A. Boyanov[1], Dimitar A. Tabakov[2], Radina S. Ivanova[2] and Kalin N. Vidinov[3]*

[1]*Clinic of Endocrinology, Metabolism, University Hospital Alexandrovska, Bulgaria*

[2]*Clinic of Endocrine Surgery, Clinical Center of Endocrinology and Gerontology, Medical University of Sofia, Bulgaria*

[3]*Laboratory of Clinical Pathology, Clinical Center of Endocrinology and Gerontology, Medical University of Sofia, Bulgaria*

Abstract

The incidence of Thyroglossal Duct Cyst Carcinoma (TGDCC) is very low usually being discovered incidentally after surgical cyst removal. We describe a case of a 17-year old girl with a midline neck cyst interpreted as a Thyroglossal Duct Cyst (TGDC). After the ultrasound examination the patient was directly referred for surgery. The histology revealed the presence of a papillary carcinoma within the cystic wall. The patient was classified as being low-risk for recurrent disease and no further surgical procedures were offered. One year after the operation the thyroid gland appeared normal with no suspicious lymph nodes present. We discuss the role of pre-operative fine-needle aspiration biopsy and ancillary imaging modalities (computed tomography etc.). The risk/benefit ratio of radical approaches such as total thyroidectomy with or without radioiodine ablation is also discussed. In conclusion, harmless thyroid pathology may harbor neoplastic foci, which highlights the role of extended pre-operative assessment.

Keywords: Thyroglossal duct cyst; Papillary carcinoma; Histology

Introduction

Thyroglossal Duct Cysts (TGDC) is by far the most common form of congenital midline neck cysts. They account for more than two thirds of the congenital neck masses [1]. Thyroglossal Duct Cyst Carcinoma (TGDCC) has been found in 0.7% to 1.5% of the TGDC [2-4]. Since the initial description by Brentano in 1911 [5], many authors reported cases and case-series of TGDCC [6-15]. A total of 250-300 cases of TGDCC have already been reported in the literature. The pre-operative and postoperative approaches may vary a lot, and a consensus on the management of those malignancies is still lacking. A large recent meta-analysis including 164 patients with TGDCC reported that 73.3% of the cases were found post-operatively with the most common pathology being papillary cancer (92.1%) [16]. However pre-operative assessment by Fine-Needle Aspiration Biopsy (FNAB) or in some cases by Computed Tomography (CT) is highly advocated [17]. Management of TGDCC is still a matter of debate [4,19], with the Sistrunk's operation being in the basis of surgical treatment [20,21]. Thyroidectomy with or without lymph node resection and adjuvant metabolic therapies remain also as treatment options.

Case Presentation

This is the case of a 17-year old female complaining with a painless midline neck mass. The patient noticed the mass growing during the last few months with no problems caused by swelling, and no voice degradation being apparent (e.g. hoarseness). There were no symptoms or signs suggestive of thyroid dysfunction. On palpation the midline neck mass was firm although elastic and measured around 3 cm in diameter with an ellipsoid shape moving along in the act of gulping. No enlarged lymph nodes were palpable. There was no family history of thyroid cancer or prior neck irradiation. The ultrasound images of the midline neck mass are presented in Figure 1. The thyroid gland had a normal appearance on US, and no thyroid nodules or suspicious lymph nodes were found. No other diagnostic procedures were performed and the patient was directly referred for surgery. The neck mass was removed with the preservation of the neck muscles and hyoid bone. The cyst had a well-defined capsule with no invasion or attachment to adjacent structures noted. No suspicious lymph nodes were found and removed. On pathology the cyst weighed 7.5 gm and measured 3 cm × 2 cm, well defined by thick fibrous capsule. On microscopy there was a classical

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Correspondence:
Mihail A. Boyanov, Clinic of Endocrinology and Metabolism, University Hospital Alexandrovska, 1, G. Sofiyski str., Sofia 1431, Bulgaria, Fax: +359 29230779; E-mail: mihailboyanov@yahoo.com

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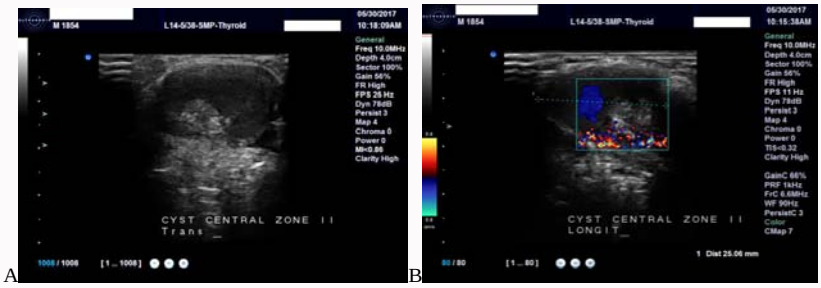

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Figure 1: Basal ultrasound images of the TGDCC: A) transversal view (gray-scale US); and B) Longitudinal view (Color Doppler US). The solid component with multiple hyper echoic spots can be easily seen. The Doppler US does not reveal intra-nodular vascularization, but a slow movement of the cystic fluid.

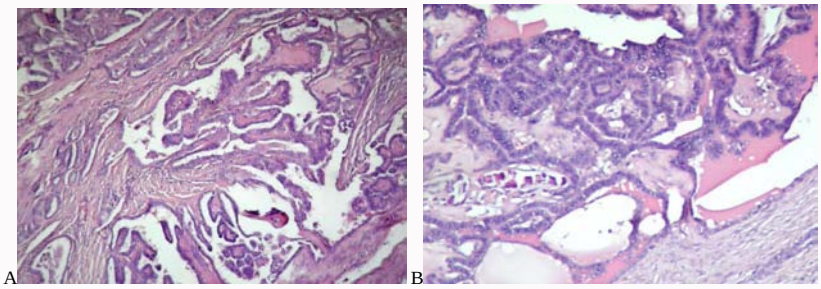

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Figure 2: (A-D) The histology of TGDCC in the 17-years old female is shown. (A-C): The presence of cystic papillary carcinoma with papillary cell structures and nuclear features typical for papillary thyroid carcinoma is displayed -clear nuclei with irregular contour, some with nuclear grooves or inclusions (HE, x 40, 60). (D): The fibrous capsule of the cyst with invasion by the papillary carcinoma can be clearly seen (HE, x 60).

papillary carcinoma, with invasion of cyst capsule and focal squamous cell metaplasia. The histology findings are presented in Figure 2. The tumor was staged pT2Nx. The post-operative US follow-up revealed a completely normal thyroid gland, no cyst residues or suspicious lymph nodes. It was decided for a wait-and-see approach - 10 months later the thyroid gland and neck region appeared clear on ultrasound (Figure 3). No symptoms or signs of disease recurrence were evident until the time of this publication.

Discussion and Comments

This is a case of incidentally discovered TGDCC in an adolescent girl. The papillary carcinoma was discovered on histology with the surgical approach being minimally invasive. A watch full waiting strategy was preferred to possible thyroidectomy and lymph node dissection. The remaining thyroid gland was of normal appearance. At least 3 questions remain open when discussing this case:

1. Is direct referral for surgery the best option? What is the place of FNAB?

2. Are there other imaging modalities beyond ultrasound that would have provided additional information?

3. Is minimally invasive surgery (preserving even the hyoid bone) an adequate approach?

As seen on Figure 1 (preoperative ultrasound image of the TGDC) there was a solid component in the cyst that did not show any Vascularity on Doppler ultrasound. A FNAB of this component might have revealed its malignant nature and lead to a more aggressive surgical approach. In the description of a case of TGDCC the authors emphasized the need for preoperative FNAB [17]. The cytological appearances of this carcinoma as well as the possible differential diagnoses are extensively discussed. The authors however admit that failure in achieving an accurate diagnosis of papillary carcinoma arising in TGDC is not uncommon [17]. A publication, including a large case series (14 cases with a neoplasm in a TGDC), reported the various methods for preoperative assessment [22]. Ten patients had nuclear scanning of the thyroid bed, 5 underwent FNAB. Of those five patients the diagnosis of a papillary carcinoma was made in only 3 cases [22]. In another series of five patients it was come to the conclusion, that TGDC in the adult population must be studied through FNAB with a frozen section in cases with indeterminate or unavailable cytology [19]. However, a meta-analysis including 164 patients revealed that 73.3% of cases were found on final pathologic analysis [16].

Another possibility for further imaging is the CT-examination [15,18]. It can reveal the solid component of the cyst (mural nodules) with possible calcifications throughout the mass as well as the adjacent structures and lymph nodes. The thyroid should also be included in the CT-scan with special attention from artifacts from the shoulders that can partially obscure the gland [18]. In our case, a CT-scan would have urged a more aggressive surgical approach. However, the question remains open, how radical the surgical approach in such cases should be? The Sistrunk's operation is the recommended first line treatment [4,16,19]. There is also space for robot-assisted surgical removal of the cyst and thyroid gland [21]. It is unclear, whether incidentally discovered TGDCC should always be followed by total thyroidectomy. Older publications state that the Sistrunk's operation might be adequate for most patients with incidentally diagnosed TGDCC in the presence of a normal thyroid gland [4]. They found that the addition of total thyroidectomy did not significantly change the outcome [4]. Recent analyses advocate thyroidectomy among patients ≥ 45 years of age and individuals with aggressive disease [16]. Others believe that thyroidectomy should be performed in all TGDCC patients [23]. Their arguments include the possibility that TGDCC might be a metastasis from an occult primary thyroid carcinoma [23]. Additionally, one might argue whether the

availability of specific mutations (like BRAF) might predispose to multiple PTC. In a study examining mutated BRAF (V600E) the authors came to the conclusion that the majority of TGDCC most likely developed as a primary malignancy from a thyroid remnant [24]. In this study the presence of V6ooE BRAF mutations did not change the outcome of disease-free survival [24]. Another approach would be to apply the risk staging and treatment algorithm for thyroid nodules as proposed by the American Thyroid Association [25].

In conclusion, this report presents a case demonstrating that the initial assessment of TGDC should be complex (ultrasound, US-guided FNAB, CT if needed, molecular and genetic analyses) bearing in mind the rare possibility for a thyroid carcinoma within the cyst. The treatment also provides challenges as the risk/benefit ratio of implementing more radical approaches (total thyroidectomy, radio-iodine ablation etc.) should always be carefully weighed.

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