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Introduction

Thyroglossal duct cysts (TGDC) account for more than two thirds of congenital neck masses [1]. Thyroglossal duct cyst carcinoma (TGDC) has been found in 0.7–1.5% of all cases of TGDC [2, 3]. Since its initial description in 1911, a total of 250–300 cases of

TGDCC have been reported as cases and case series of TGDCC [4–9]. A recent large meta-analysis including 164 patients with TGDCC reported that 73.3% of the cases were found post-operatively [10]. However pre-operative assessment by fine-needle aspiration biopsy (FNAB) and sometimes by computed tomography (CT) is highly advocated [11, 12]. Management of TGDCC is still a matter of debate, with Sistrunk's operation being the basis of surgical treatment [13–15].

Case presentation

A 17-year-old female presented to our hospital with a painless midline neck mass. The patient noticed the mass growing for the last few months with no problems with 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 but elastic and measured approximately 3 cm in diameter with an ellipsoid shape and moving during the act of swallowing. 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 g, measured 3 × 2 cm, and was well defined by a thick fibrous capsule. On microscopy there was a classical papillary carcinoma, with invasion of the cyst capsule and focal squamous cell metaplasia. The histology findings are presented in Figure 2. The tumour was staged pT2MxNx. The post-operative US follow-up revealed a completely normal thyroid gland, and no cyst residues or suspicious lymph nodes. A wait-and-see approach was adopted. Ten months later the thyroid gland and neck region appeared normal on ultrasound. No symptoms or signs of disease recurrence have been evident until the time of this publication.

The data described in this case report were gathered retrospectively. The management of the patient was part of the routine clinical work-up. The subject and her parents have given their written, informed consent to publish the case.

Discussion

We present a case of TGDCC in an adolescent girl incidentally discovered on histology with the surgical approach being minimally invasive. At least three questions remain open for discussion:

1. Is direct referral for surgery the best option? What is the place of FNAB?
2. Are there other imaging modalities beyond ultrasound that could have provided additional information?
3. Is minimally invasive surgery (preserving even the hyoid bone) an adequate approach?

As seen in Figure 1, 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 led to a more aggressive surgical approach. In another description of a case with TGDCC the authors emphasised the need for preoperative FNAB [11]. Another publication, including 14 cases with a neoplasm in a TGDC, assessed the various methods for preoperative assessment [16]. In the five patients who underwent FNAB the diagnosis of a papillary carcinoma was made in only three cases [16]. Another report on a series of five patients came 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].

Another possibility for further imaging is CT-examination [9, 12]. 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. In our case, a CT scan would have urged a more aggressive surgical approach.

However, the question remains open: how radical in such cases should the surgical approach be? Sistrunk's operation is the recommended first-line treatment [5, 13]. There is also space for robot-assisted surgical removal of the cyst and thyroid gland [15]. It is unclear whether incidentally discovered TGDCC should always be followed by total thyroidectomy. Older publications found that the addition of total thyroidectomy did not significantly change the outcome [1]. Recent analyses advocate thyroidectomy among patients ≥ 45 years of age and individuals with aggressive disease [10]. Others believe that thyroidectomy should be performed in all TGDCC patients [17]. Their arguments include the possibility that TGDCC might be a metastasis from an occult primary thyroid carcinoma [17].

In conclusion, this report presents a case demonstrating that the initial assessment of TGDC should be complex (ultrasound, US-guided FNAB, etc.), bearing in mind the rare possibility for a thyroid carcinoma within the cyst. The treatment also provides challenges because the risk/benefit ratio of more radical approaches should always be carefully weighed.

Disclosure statement

The authors have no conflicts of interest to disclose

Statements of ethics

The research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki. Data were collected retrospectively.

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Competing interests

None declared.

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Figure 1. Basal ultrasound images of the thyroglossal duct carcinoma (TGDC) — transversal view (greyscale US). The solid component with multiple hyperechoic spots can easily be seen. The Doppler US did not reveal intra-nodular vascularisation, but a slow movement of the cystic fluid



Figure 2. The histology of thyroglossal duct cyst carcinoma (TGDC) in the 17-year-old female is shown. **A.** The fibrous capsule of the cyst with invasion by the papillary carcinoma can be clearly seen (HE, $\times 40$). **B.** 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, $\times 60$)



