Thyroglossal duct papillary carcinoma with squamous metaplasia in a 12-year old female and review of literature

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A B S T R A C T

We are reporting a case of a 12-year old African-American female who presented with a six-month history of a painless, enlarging, midline neck mass. Imaging demonstrated a 4-cm solid mass with cystic components and calcifications. Differential diagnosis included dermoid tumor, teratoma, sarcomatous lesion, thyroglossal duct cyst carcinoma, and ectopic thyroid tissue with malignant transformation. Preoperative thyroid studies were unremarkable. Patient underwent Sistrunk procedure and cervical lymph node dissection. Pathology reported a diagnosis of papillary carcinoma with squamous metaplasia arising from a thyroglossal duct cyst with three of five positive lymph nodes. Post-operative thyroid studies and imaging were unremarkable. After literature review on pediatric cases of thyroglossal duct carcinomas, the decision was made to forgo thyroidectomy and opt for close follow-up. Thyroglossal duct carcinoma is a rare pediatric tumor with approximately 26 cases reported in the literature. Best practice for the extent of surgical resection and post-operative management is still evolving as we gather data on long-term outcomes.

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The thyroid gland descends from the foramen cecum, located at the base of the tongue, to the anterior neck below the thyroid cartilage along a thyroglossal duct tract. This tract disappears by the 10th week of gestation; however, incomplete atrophy of the tract leads to a persistent remnant that can lie anywhere along the tract between the tongue and thyroid gland, and it may contain ectopic thyroid tissue [1]. Thyroglossal duct (TGD) cyst is the most common congenital cyst in neck and is the second most common cause of a midline neck mass; first being lymphadenopathy [2]. TGD remnant may be present but not detected clinically by 7% of the population are believed to have subclinical TGD remnants [3]. In the pediatric population, the mean age of presentation is six years of age with enlargement of the TGD remnant occurring secondary to hypothyroidism (surge in thyroid stimulating hormone (TSH) stimulates ectopic thyroid tissue within the duct remnant causing it to enlarge), puberty, pregnancy, upper respiratory tract infection, or radiation therapy [4].

In the general population, approximately 1% of TGD cysts have evidence of carcinoma [5]. Including our case, there have been 27 case reports of adolescents with a thyroglossal duct cyst carcinoma (TGDCa) with a mean age of diagnosis between 12 and 13 years of age (12.7) [4]. All of the TGDCa have presented as asymptomatic masses with patients having no complaints of dysphagia, odynophagia, hoarseness, dyspnea, or symptoms of hyper- or hypothyroidism. In the pediatric literature (under 18-years of age), all the carcinomas have been discovered after surgical excision and have been papillary carcinoma with three cases being a mixed papillary-follicular carcinoma. There is a potential for local spread as 45% had capsular invasion and 23% had local invasive disease involving musculature, multi-focal disease, and nearby blood vessels [4]. However, distant metastases are quite rare. In the adult population, 1.3% of papillary carcinoma of TGD cyst present with evidence of distant metastasis and in the pediatric population there has been only one documented case of distant metastatic disease [6]. In this paper, we report a case along with our workup and treatment plan of a 12-year old female who presented with a TGDCa.

1. Case report

A 12-year old African-American female presented with a six-month history of an enlarging, painless, midline neck mass. She denied any symptoms of dysphagia, odynophagia, dyspnea,
Fig. 1. Sagittal computed tomographic image of the neck. Blue arrow: predominately solid mass with cystic components and micro-calciﬁcations. Yellow arrow: hyoid bone, red arrow: thyroid gland.

Fig. 2. Axial computed tomographic image of the neck. Blue arrow: predominately solid mass with cystic components and micro-calciﬁcations.

The mass was predominantly solid with cystic change surrounded by stromal stromal metaplasia and minimal chronic inﬂammation. There was focal involvement of the nearby skeletal muscles. Three of ﬁve (3/5) lymph nodes were positive for metastatic carcinoma. The hyoid bone was free of carcinoma and the resection margins were free of tumor.

2. Discussion

TGDCa is quite rare and a difference of opinions in best management practices still exists among experts. Management of these carcinomas involves surgical excision via a Sistrunk procedure. However, whether an additional operation for cervical lymph node dissection and thyroidecotomy along with radioactive iodine ablation is indicated has yet to be determined. In the adult population, thyroidecotomy is deﬁnitely indicated as a link between TGDCa and thyroid cancer has been identiﬁed, as 25–33% of adults with TGDCa have evidence of carcinoma in their thyroid gland [7,8]. However, in the pediatric population, thyroidecotomy has not been proven to improve prognosis in patients with TGDCa as no link between pediatric TGDCa and thyroid carcinoma has been identiﬁed. Of the previous 26 cases, 13 have undergone total thyroidecotomy, none of which showed any evidence of thyroid carcinoma [9]. Unfortunately, there are no large randomized-control trials to support one side over the other. In our patient, the decision was made to forgo thyroidecotomy and opt for close follow-up since no link has been established between TGDCa and a thyroid malignancy and because there is a very low risk of distant metastasis. We felt it was best to not subdue our patient to the morbidity of a second operation and the need for life-long thyroid hormone supplementation. We will continue to monitor for thyroid involvement with close follow-up at 6-month intervals with imaging and thyroid function tests. 3- and 6-month follow-ups were unremarkable.

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References