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Case Report

Thyroglossal duct papillary carcinoma in a 15-year old female and review of pediatric cases of thyroglossal duct carcinoma

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ABSTRACT

Thyroglossal duct carcinoma is rare, occurring in approximately 1% of thyroglossal duct cysts. Excluding this case report there have been 25 cases of adolescent thyroglossal duct carcinoma reported in the English literature thus far. Most of the pathology reported has been papillary or follicular carcinoma, leading to the question of whether or not to perform concurrent thyroidectomy. Based on our review of the pediatric cases of thyroglossal duct carcinoma we elected not to perform a concurrent thyroidectomy and recommend close follow-up to monitor for signs of future thyroid involvement.

1. Introduction

Thyroglossal duct cysts (TGDC) are the embryologic remnant of the descending tract of the thyroid gland as it courses from the foramen cecum at the base of the tongue to its final resting position in the neck. Failure of the tract to fully involute can result in a retained cyst with associated thyroid tissue in the stroma. The cyst is lined with either pseudostratified columnar or squamous epithelium [1]. Remnants of the thyroglossal duct occur in roughly 7% of the population [2]. Due to the embryologic development, physical exam findings of a TGDC consist of a midline mass that moves with swallowing and deglutition.

Malignancy of the thyroglossal duct remnant is rare, occurring in approximately 1% of cases of TGDC [3,4]. Approximately 90% of thyroglossal duct carcinomas (TGDCa) are either of papillary or follicular origin, while the remainder are either squamous cell carcinoma (SCC), anaplastic carcinoma, Hürthle cell carcinoma, or undifferentiated unspecified adenocarcinoma [5,6]. Papillary and follicular cell carcinomas develop from ectopic thyroid tissue in the surrounding duct remnant; SCC develops from the epithelial lining of the cyst itself [7]. Medullary carcinoma does not develop in thyroglossal duct remnants because it arises from parafollicular C cells which are of neural crest origin.

TGDCa in children is even more rare than in adults, with twenty-six cases reported in the English literature including this report [8–12]. Although treatment recommendations have been proposed for adults, limited guidance exists regarding treatment in children. This paper presents the case of a 15-year-old female with TGDCa, her diagnostic work up, and her treatment plan. In addition, we evaluate the cases of pediatric TGDCa reported in the literature and discuss treatment options.

2. Case report

A 15-year-old female was seen in the office with a painless submental mass first noticed 2 months prior. The patient denied dysphagia, odynophagia, dyspnea, hoarseness, history of radiation exposure or family history of thyroid carcinoma. She did not have any symptoms of hyper- or hypothyroidism. Clinical examination revealed a firm, nontender, midline neck mass approximately 4 cm × 4 cm that was adherent to the hyoid bone and moved with swallowing. There was no fluctuance or overlying skin changes and no thyroid nodules or cervical lymphadenopathy was noted.

The patient was referred to our clinic with computed tomography (CT) of the neck and I-131 scan. Neck CT without contrast revealed a lobulated, partially calcified mass attached to the anterior hyoid bone with extension from the base of tongue and deep tongue musculature (Fig. 1). I-131 scan showed normal uptake in bilateral thyroid lobes without any hot or cold nodules. Ultrasound guided fine-needle aspiration cytology (FNAC) revealed low-grade atypical follicular epithelial cells with scattered intranuclear pseudoinclusions and nuclear grooves highly

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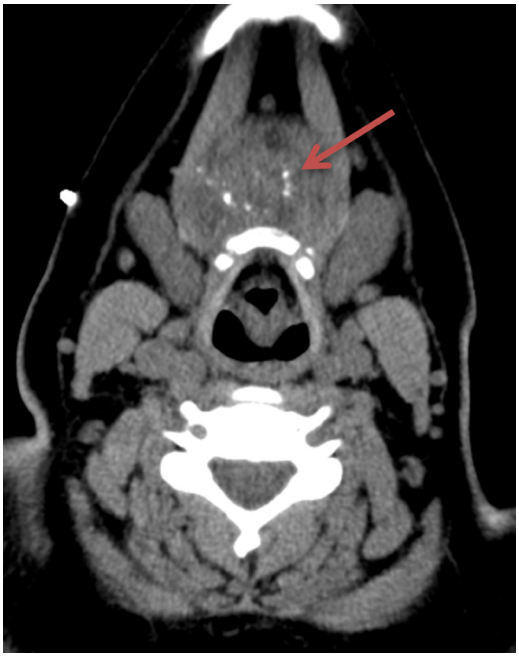

Fig. 1. Axial image of submental mass adherent to hyoid bone with calcifications.

suspicious for papillary carcinoma, possibly arising from a thyroglossal duct cyst.

The patient was scheduled for surgery and a Sistrunk procedure with wide local excision was performed under general anesthesia without complication. The gross pathologic specimen revealed a 2.3 cm × 2.0 cm × 2.0 cm red-tan mass with ill-defined margins invading the skeletal muscle and hyoid bone. Cross-section of the mass showed a soft and friable center with calcifications present. Associated bland lymphoid tissue was suggestive of a thyroglossal duct remnant. Microscopic findings of psammoma bodies and cells with large and irregular nuclei containing prominent grooving and intranuclear inclusions (Figs. 2 and 3) led to the diagnosis of papillary carcinoma. Three lymph nodes were evaluated and found to be negative for local lymphatic invasion.

3. Materials and methods

A PubMed search was performed in August 2012 for "thyroglossal duct 'AND' carcinoma 'OR' papillary". Articles were reviewed and selected if patients were from 0 to 18 years old. A previous review of pediatric TGDCa was performed in October 2003 [8], narrowing our search to October 2003–August 2012. Only articles in English were included in our analysis.

Information was compiled regarding patient age, gender, pathology, co-existent thyroid malignancy, nodal involvement, and local involvement. Results were organized (see Table 1) and analyzed.

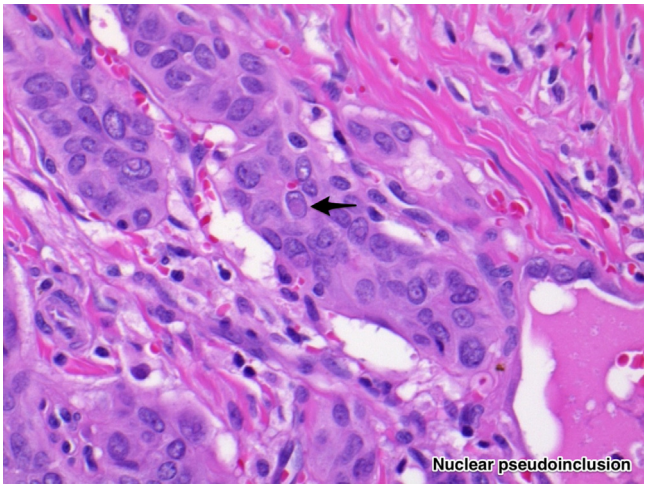

Fig. 2. Invasive tumor cells at 40× magnification. Tumor cells show optically clear, large and irregular nuclei with intranuclear inclusions.

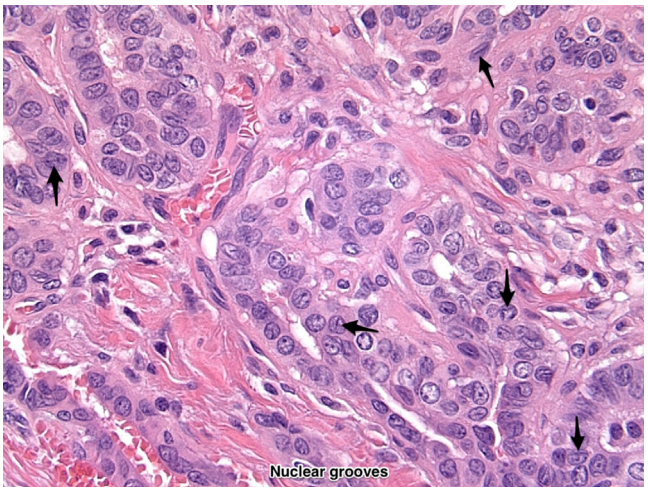

Fig. 3. Invasive tumor cells at 40× magnification. Tumor cells show irregular nuclei with prominent grooving.

4. Results

Including this case, there have been twenty-six cases of childhood TGDCa reported in the English literature. Another case was discovered in our search, but the article was in Spanish and thus not included in our analysis [27]. Of the cases analyzed, the mean and median age at diagnosis was 12.7 and 13.5 years, respectively. Fifteen of the subjects were female and ten were male; the gender of one of the subjects was not included in the study. Twenty-two of the cases were papillary carcinoma, three were mixed papillary–follicular, and no information was given regarding tumor pathology for one patient. Thirteen of the patients underwent total thyroidectomy, none of which showed malignant involvement of the thyroid gland. The number of patients with local lymph node involvement was 5/10 (50%), one of which had positive node involvement 2 years after initial surgery. Of the cases commenting on local tumor invasion, 9/11 (82%) were found to have involvement of the cyst wall, surrounding muscle, or vasculature.

5. Discussion

Thyroglossal duct carcinoma is a rare entity, especially in children. The dilemma arises regarding treatment of TGDCa in children. Several suggestions have been made for treatment in adults. Dzodic et al. reviewed the treatment of 12 patients (9 females and 3 males, mean age of 40.6 at diagnosis, age range 27–63) with TGDCa. Eleven of the patients underwent total thyroidectomy and one limited thyroidectomy. Three of the 12 patients (25%) had thyroid carcinoma. Those with thyroid carcinoma had subsequent level VI neck dissection, which was positive for nodal involvement in 2/3 cases. Routine level I neck dissection was positive in 6/11 patients. Dzodic et al. recommend Sistrunk procedure with thyroidectomy and level I neck dissection, level II–IV dissection if lymph node biopsy positive, and level VI dissection if concurrent thyroid carcinoma [28].