Canon
Journal Of Medicine
www.Canonjm.com
DOI 10.30477/CJM.2019.81660
Case Report

Thyroglossal Duct Cyst Carcinoma in Children: Case Report and Review of Management

Zahra Sarafraz[1*] , Seyyed Ali Musavi[2] , Mansour Moghimi[3]

1- Assistant professor, Department of Otolaryngology, Shahid Beheshti University of Medical Sciences, Tehran, Iran

2-Assistant professor, Department of Otolaryngology, Shahid Sadoughi University of Medical Sciences, Yazd, Iran

3-Assistant professor, Department of Pathology, Shahid Sadoughi University of Medical Sciences, Yazd, Iran

*Corresponding author: **Zahra Sarafraz**

Address: Assistant professor, Department of Otolaryngology, Shahid Beheshti University of Medical Sciences, Tehran, Iran

E-mail: zahra.sarafraz@yahoo.com

Phone:+989125071121

Recive Date: 11 January 2019, Revise Date: 14 January 2019, Accept Date: 15 January 2019

ABSTRACT

Introduction: A thyroglossal duct cyst (TDC) is the most common congenital neck mass. Seventy five percent of midline neck masses in children are TDCs. Malignant transformation occurs in 1% of benign cysts. The most common is papillary thyroid carcinoma (80 %). It usually presents in the third or fourth decade of life and is more common in women.

Case presentation: In this case report, we present the case of a 5-year-old boy with thyroglossal duct cyst carcinoma and review of management.

Conclusion: PTC occurs in TDC rarely, but the probability should be considered before surgery, especially in high risk groups. It is advisable to choose more conservative management plans for children to preserve their quality of life and prevent iatrogenic complications.

Keywords: Thyroglossal Duct Cyst; Papillary Thyroid Carcinoma; Neck Mass.

INTRODUCTION

A thyroglossal duct cyst (TDC) is the most common congenital neck mass. TDC includes 75 % of midline neck mass in children. This anomaly is a result of regression failure of the tract between the thyroid gland and the root of tongue at the sixth week of life. Malignant transformation occurs in 1% of these benign cysts. It usually presents in the third or fourth decade of life and is more common in women than men. It is rarely seen in people under 14 years of aage. The diagnosis is usually made by histopathologic examination after neck mass excision. The most common (80%) malignant tumor is papillary thyroid carcinoma (PTC), followed by mixed papillary-follicular carcinoma (8%), squamous cell carcinoma (6 %), follicular carcinoma (3 %), adenocarcinoma, and unidentified tumors (3 %) [1-3].

The medullary carcinoma has not been seen in the thyroglossal duct cyst [4]. The prognosis of PTC is excellent in the thyroglossal cyst, compared to other rarer types. There is a controversy about thyroglossal duct carcinoma management, and there are different opinions about the value of the Sistrunk procedure (which involves a complete cyst resection and resection of the central part of hyoid bone), with or without total thyroidectomy [5]. In this study, we introduce a 5-year-old Iranian boy with thyroglossal duct cyst carcinoma and describe the management of his case.

CASE PRESENTATION

A 5-year-old Iranian boy had a right lateral neck mass. The size of mass was 30*20 mm, without any inflammatory features (redness, tenderness, or warmness).

The mass used to move with swallowing. It was gradually grown over a two-year period. The child had no respiratory distress, hoarseness, odynophagia, or dysphagia. There was no other finding on the neck examination regarding lymphadenopathy or thyroid nodule. Parents reported no previous infection in the neck (figure1). Ultrasonography showed a complex cystic mass with solid component. The mass was 32*24 mm. No nodule was detected in thyroid sonography, and neck lymphadenopathy was negative. Fine needle aspiration (FNA) was performed and squamous cells and cyst macrophages were seen. A neck CT scan with contrast showed a well-delineated cystic lesion with a small solid component and its relation to hyoid bone. A standard Sistrunk procedure was performed and his permanent pathology revealed a white, firm, encapsulated mass of 30*20mm size with fronds of tissue with thin fibrovascular cores. The fronds had an overall papillary pattern. Papillae were lined by cuboidal cells, and the nuclei had clear chromatin. There were longitudinal grooves that suggested papillary thyroid carcinoma (PTC) in TDC. There were no features of invasion in microscopic evaluation. (figure2). A family history for PTC was negative. There was no history of radiation exposure. A close follow up was performed with neck sonography every three months. After five years, the patient has seen no recurrences or symptoms or features.

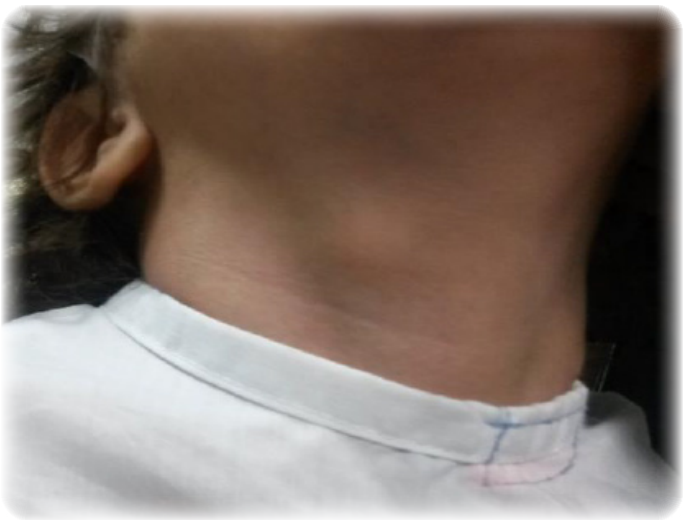

Figure1. A 5-year-old boy with a right lateral neck mass

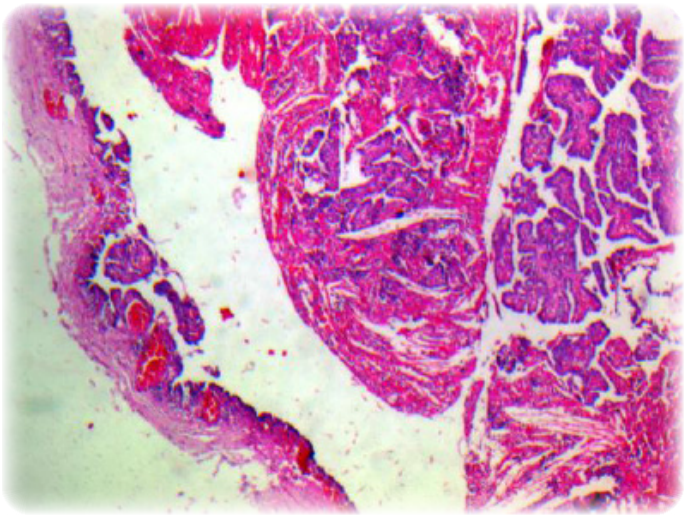

Figure2. Papillary pattern in thyroglossal duct cyst

DISCUSSION

A thyroglossal duct cyst (TDC) is the most common congenital neck mass. Malignant transformation occurs in 1% of these benign cysts. In pediatric literature review the most common malignant tumor is PTC (80%) [1]. Three patients presented with a mixed papillary–follicular carcinoma and 2 patients with no information regarding tumor pathology [1, 2].

There are two theories to explain the thyrogenic origin of thyroglossal duct carcinoma (TGDC); first is a de novo tumor. An absence of a medullary carcinoma in the TGDC as it arises from parafollicular cells supports this theory. The second is second metastasis from thyroid PTC [6]. It is usually presented in the third or fourth decade of life. It is very rare in children and almost all studies of TGDC among them are just case reports [7]. In almost all pediatrics with complete clinical presentation in literature, the mass was asymptomatic, except for a 13-year-old girl with a “mildly tender” mass [2]. However, our case was a 5-year-old boy with an asymptomatic mass.

For TDC carcinoma, correct and complete management especially in children is unclear. Most authors acknowledge the value of at least a Sistrunk procedure after TDC diagnosis because of the high incidence of infection and the possibility of malignancy that increases by age.

Some authors recommend a thyroidectomy on the basis of risk. High-risk characteristics are defined as:

• Patient age greater than 45

• History of radiation exposure

• Tumor in the thyroid on imaging
• Clinical or radiological nodes
• More than 1.5 cm in diameter (Although the size of the neck mass with TGDC is not an indicative factor for malignancy in children) [2].
• Cyst-wall invasion
• Positive margins on histopathologic examination [1].

Our case showed none of these risk factors.

N. Balalaa et al. considered histopathology for operation type selection. They recommend Sistrunk operation alone for squamous cell carcinoma, but total thyroidectomy for differentiated thyroid carcinoma [6]. Some authors recommend the Sistrunk procedure with total thyroidectomy because they believe that PTC is multi-focal and has lymphatic invasion. With a total thyroidectomy, we can do correct follow up [7]. In some studies, the cure rate for papillary thyroid carcinoma (PTC) in TDC has been reported as 95 percent when treated by a Sistrunk procedure alone. Furthermore, in a recent publication, the survival rate was reported as 100 percent when treated by Sistrunk and total thyroidectomy with lymph node dissection [1, 7].

Management in children includes several considerations. Twelve thyroidectomies have been reported in the 22 pediatric patients with TGDC. All of the resected thyroid specimens were free of carcinoma [2]. Distant metastases are very rare in TGDC, and have been described in only 1.3% of patients with papillary TGDC, mainly in the adults. Only one pediatric patient was reported to have lung metastases due to TGDC. The prognosis of TGDC in the pediatric population was favorable, as evidenced by the fact that only 1 patient had died, 8 hours after surgery (may be because of the surgery complications) [2].

Well-differentiated thyroid carcinoma and TGDC have favorable prognosis. The 5- and 10- year overall Kaplan-Meier survival of adults was 100% and 95 %, respectively.

None of the patients in pediatric study had recurrence in the surgical bed. There were no disease-related deaths reported in any of the children. Total thyroidectomy and cervical node dissection have not any significant impact on outcome. Because of good prognosis in children and high morbidity and negative effects on quality of life after thyroidectomy including operative and postoperative complications (3%–5% incidence of hypocalcemia and 1%–2% incidence of recurrent laryngeal nerve injury) and lifelong thyroid hormone dependency, Sistrunk with close follow up is recommended as the standard procedure [2]. In five years, our case showed no symptoms or signs of recurrence.

CONCLUSION

PTC occurs in TDC rarely, but the probability should be considered before surgery, especially in high risk groups. We should select an individualized treatment plan for each patient. It is advisable to choose more conservative management plans for children to preserve their quality of life and prevent iatrogenic complications.

Authors' Contributions:

ZS presented the case and drafted the manuscript. SAM helped in manuscript drafting and data presentation. MM helped in manuscript drafting and designed the study. All authors have approved the final version of manuscript.

Conflict of Interest Disclosures:

There are no conflict of interest in terms of the present manuscript.

Ethical approval/Consideration:

A written informed consent form was taken from patient's guardian for reporting his case. All the personal information remained anonymous.

REFERENCES

1. Choi YM, Kim TY, Song DE, Hong SJ, Jang EK, Jeon MJ, et al. Papillary thyroid carcinoma arising from a thyroglossal duct cyst: a single institution experience. Endocrine journal. 2012;60(5):665-70.

2. Peretz A, Leiberman E, Kapelushnik J, Hershkovitz E. Thyroglossal duct carcinoma in children: case presentation and review of the literature. Thyroid. 2004;14(9):777-85.

3. Cevik c. YM, Goke C. Papillary Carcinoma Aris-

ing in a Thyroglossal Cyst with Thyroid Microcarcinoma not seen by Sonography. Ind J Med Case Rep. 2013;2(1):5-8.

4. Doshi SV, Cruz RM, Hilsinger Jr RL. Thyroglossal Duct Carcinoma: Alarge Case Series. Annals of Otology, Rhinology & Laryngology. 2001;110(8):734-8.

5. Ramalingam R, Ramalingairr K, Ramesh A, Prabhu SM, Vergis M, Ahilasamy N. Papillary thyroid carcinoma in a thyroglossal cyst. Indian Journal of Otolaryngology and Head & Neck Surgery. 2003;55(4):294-5.

6. Balalaa N, Megahed M, Al Ashari M, Branicki F. Thyroglossal duct cyst papillary carcinoma. Case reports in oncology. 2011;4(1):39-43.

7. Falvo L, Giacomelli L, Vanni B, Marzollo A, Guerriero G, De EA. Papillary thyroid carcinoma in thyroglossal duct cyst: case reports and literature review. International surgery. 2006;91(3):141-6.