PRIMARY THYROGLOSSAL DUCT CYST PAPILLARY CARCINOMA: A CASE REPORT

Mutahir A. Tunio[1], Mushabbab Al Asiri[2], Khalid Hussain Al-Qahtani[3], Khalid Riaz[4]

[1,2,4] Department of Radiation Oncology, Comprehensive Cancer Center, King Fahad Medical City, Riyadh - Saudi Arabia
[3] Department of Otolaryngology-Head & Neck Surgery, College of Medicine, King Saud University, Riyadh - Saudi Arabia.
Address for correspondence:
Dr. Mutahir A. Tunio
Department of Radiation Oncology, Comprehensive Cancer Center, King Fahad Medical City, Riyadh - Saudi Arabia
E-mail: drmutahirtonio@hotmail.com
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ABSTRACT

A Thyroglossal duct cyst (TGDC) is the most frequent congenital cervical anomaly (~70% in children and 7% in adults) in the development of the thyroid gland. However, incidence of carcinoma arising from a TGDC is extremely rare (≤1%) and is diagnosed during the pathologic examination of thyroidectomy specimen. To date, only fewer related cases have been reported to the literature. Among those cases, only few were diagnosed as primary TGDC papillary carcinoma. Here in, we present a case of 25- year old woman with primary TGDC papillary carcinoma identified after sistrunk surgery. Later on, patient underwent total thyroidectomy, bilateral neck dissection and adjuvant radioactive iodine ablation. The patient was alive and without any recurrence at fifth year of follow-up after her initial diagnosis.

Key Words: Thyroglossal duct cyst, Papillary carcinoma, Rare manifestation

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INTRODUCTION

Thyroid gland starts developing in the oropharynx in the fetus and descends from the foramen cecum between tuberculum impar and cupola to its final location at the thyroid cartilage. The epithelial tract after thyroid migration is called as the thyroglossal tract; this tract usually disappears during the 5th to 10th gestational weeks. Incomplete atrophy/closure of the thyroglossal tract leads to thyroglossal duct cyst (TGDC) [1]. TGDC is the most frequent congenital cervical anomaly that occurs in 70% of children and 7% of the adult population. However, only 1% of thyroid carcinomas arise from a TGDC[2]. First case of TGDC papillary carcinoma was reported in 1911 by Brentano, et al, and after that, around 200 cases have been reported in the literature[3]. Among those cases, majority had invasion of TGDC secondary to thyroid carcinoma and only few cases are diagnosed as 'true' primary TGDC papillary carcinoma[4]. Here in, one case of primary TGDC papillary carcinoma is reported.

CASE REPORT

A 25 year old Saudi female presented in our clinic with history of painless neck swelling since last 8 months without any associated complaints (dysphagia, hoarseness of voice or weight loss). Her past medical, surgical and family history was unremarkable.

On physical examination, there was about 2 x 2 cm, non-tender soft tissue mass at mid-cervical line which was mobile with gulping. There was no palpable lymphadenopathy. The rest of systemic examination was unremarkable. Hematology, serum electrolytes, thyroid function tests (TFTs); hepatic and renal function tests were found within normal limits.

Thyroid ultrasonography showed well defined, cystic lesion of size 2.5x 2.2 x 2.0 cm with hypoechoic area. Thyroid scintigraphy revealed an increased focus of uptake in mid-anterior neck (Figure 1A & B).

The patient underwent sistrunk operation. Histopathology showed thyroglossal duct cyst with stratified squamous epithelium infiltrated by papillary cells. Immunohistochemistry (IHC) showed the positivity for thyroglobulin, CD15 and thyroid transcription factor-1(TTF-1); which confirmed the papillary carcinoma (Figure 2). A total thyroidectomy with bilateral neck dissection was performed to rule out any other focus

Figure 1: (A) Thyroid ultrasonography showing cystic lesion of size 2.5x 2.2 x 2.0 cm with hypoechoic area and (B) thyroid scintigraphy showing an intense focus in mid-anterior neck

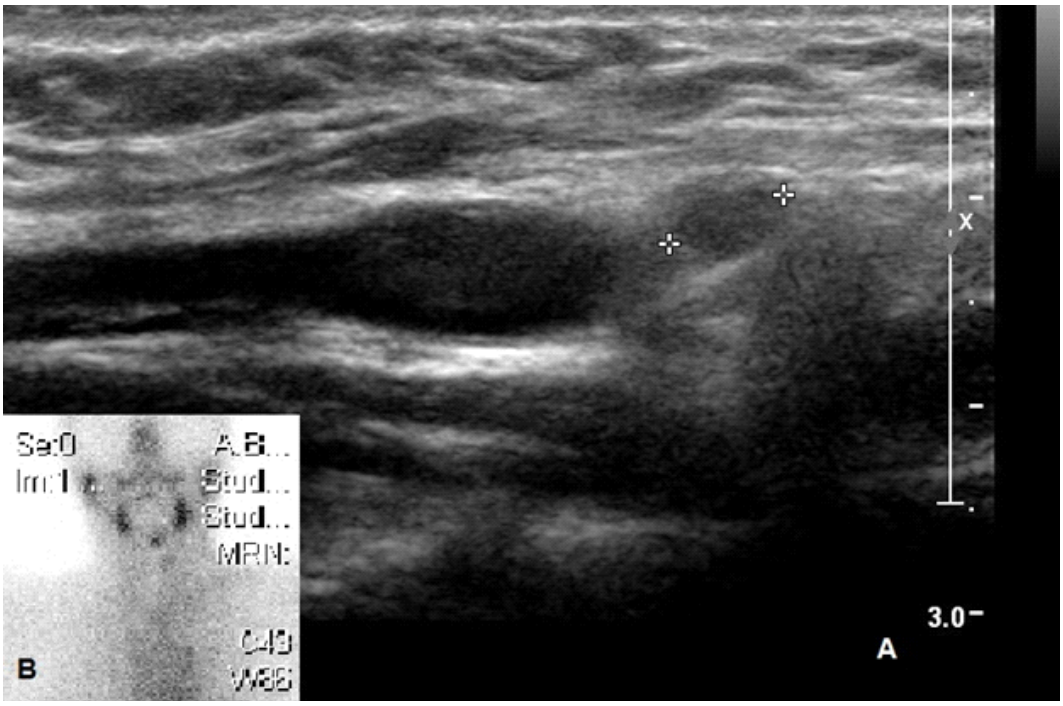
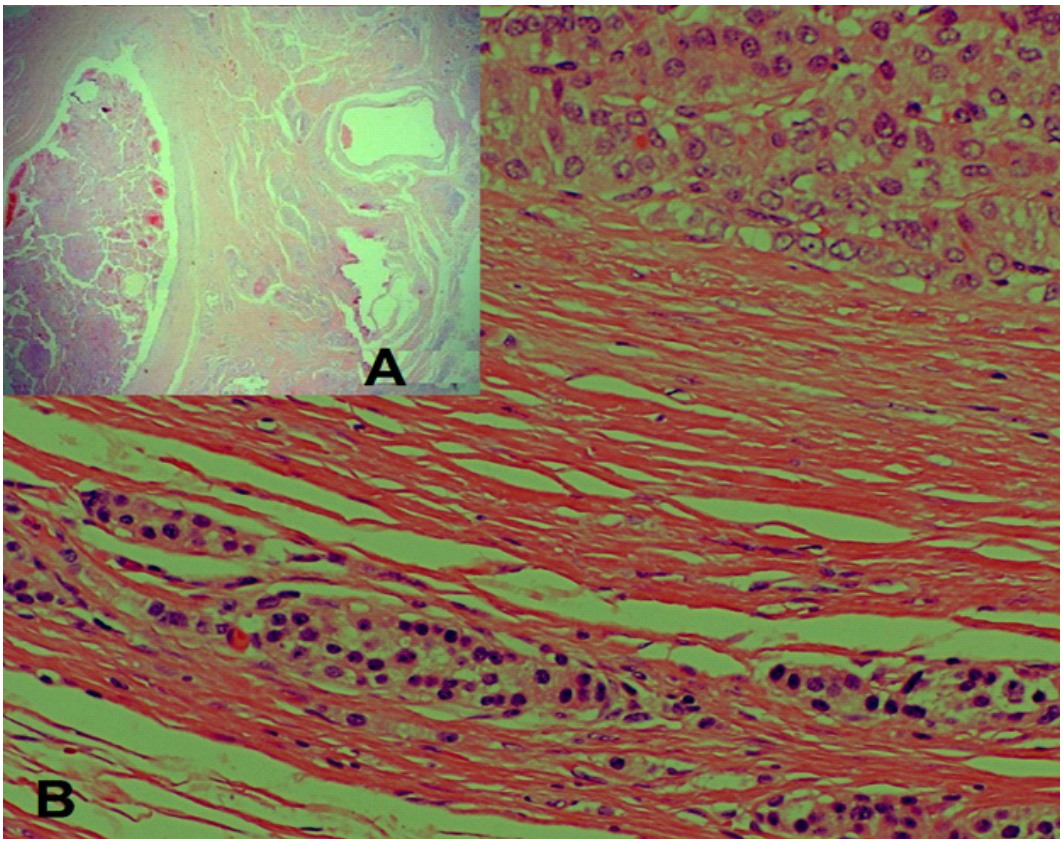


Figure 2: (A) Hematoxylin and Eosin staining showing thyroglossal duct cyst and papillary structures with follicles inside the cyst x100, and (B) Papillary cells with follicles x 200



within thyroid gland and lymph node invasion. Thyroid gland and all 16 cervical lymph nodes were negative for malignancy. The final diagnosis was made as primary thyroglossal duct cyst papillary carcinoma. Adjuvant radioactive iodine ablation (RAI) 100 mCi was given followed by thyroid suppression therapy. The patient has been following with us with any signs of recurrence or metastasis for five years.

DISCUSSION

The primary TGDC papillary carcinoma is rare manifestation with ~90% of them originating from aberrant thyroid tissue (de novo theory), in contrast to invasion of TGDC from an occult primary thyroid gland (metastatic theory)[5]. Papillary carcinomas either insular or follicular variants comprise 94%, and less than 5% are of epidermoid origin (poor prognosis)[6]. Primary TGDC papillary carcinoma remains asymptomatic in 70% of cases. Seldom, it can be revealed by dysphagia, snores or loss of weight. The diagnostic criteria of primary TGDC are; (a) histopathological identification of TGDC itself; (b) the presence of thyroid follicles in the cyst wall; (c) the existence of normal thyroid tissue adjacent to the tumor and (d) the absence of primary carcinoma in the thyroid[7].

For primary TGDC papillary carcinoma, total thyroidectomy ± neck dissection after sistrunk surgery is recommended whether the thyroid is or not involved clinically or radiologically; because of multifocal and multicentric nature of papillary carcinomas[8]. Data suggest that, adjuvant radioactive iodine ablation (RAI) followed by hormonal treatment shall be proposed only for cases in which tumors are ≥ 1 cm, multifocal or positive lymph nodes[9].

In conclusion, primary TGDC papillary carcinoma is extremely rare entity. Detailed histopathology examination is confirmatory in such cases. Total thyroidectomy is the standard treatment for such cases and adjuvant RAI shall be offered only for tumors of size are ≥ 1 cm, multifocal or with positive lymph nodes.

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