

Incidentally diagnosed papillary carcinoma in a thyroglossal duct cyst. What is the next step? A case report and review of the literature

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ABSTRACT

Thyroglossal duct cyst is the commonest congenital disorders of the thyroid gland. Rarely, it can be a malignant tumor which is picked up incidentally in postoperative histopathology. Although management is still a controversy, a careful treatment and follow up planning is necessary for the management of such condition. We present a case of a 16-year-old female patient with thyroglossal duct cyst carcinoma .

Keywords: thyroglossal cyst , papillary thyroid carcinoma , congenital thyroid disease .

تشخيص سرطان حلتمي في كيس القناة الدرقية. ما هي الخطوة التالية؟ تقرير حالة ومراجعة

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فرع الجراحة / كلية الطب / جامعة الموصل / الموصل / العراق

الخلاصة

كيس القناة الدرقية اللساني هو أكثر الاضطرابات الخلقية شيوعاً في الغدة الدرقية. في حالات نادرة ، يمكن أن يكون ورمًا خبيثًا يتم تشخيصه بالمصادفة بالفحص النسيجي بعد الجراحة. على الرغم من ان العلاج المناسب لا زال مثيرا للجدل ، إلا أن العلاج الدقيق ومتابعة التخطيط ضروري لإدارة مثل هذه الحالة. نقدم حالة مريضة تبلغ من العمر ١٦ عامًا مصابة بسرطان كيس القناة الدرقية اللسانية .

الكلمات المفتاحية: كيس قناة الغدة الدرقية ، ورم حلتمي درقي ، امراض الغدة الولاوي .

INTRODUCTION

Thyroid gland arises as a midline endodermal fold of the foregut, descends from foramen caecum in the tongue to the neck. The tract disappear in the 5th to10th week of intrauterine life, failure of involution gives rise to thyroglossal remnants¹. Thyroglossal duct cysts present as an anterior painless midline neck mass, mostly at sub hyoid region. It is commonly seen at pediatric but it can be seen in other ages as well. it is usually benign but malignancy in the cyst can be seen in 1% of thyroglossal duct cysts^{2,3}. The condition is more common in female⁴. Papillary carcinoma is the most common type (80-95%)⁵. Brentano described the first carcinoma case in thyroglossal

duct cyst in 1911⁶. The reported cases in world literature until 2017 are about 275 patients⁷. Sistrunk's operation is surgery of choice, but thyroidectomy or radioiodine therapy is still controversial and no consensus exists⁸. We present a case of a 16-year-old female who presented with a thyroglossal duct cyst in the sub-hyoid region, which was proved by postoperative histopathology as a papillary thyroid carcinoma. The report tried to evaluate the management issues involved and review of other literature and provides the use of Positron emission computerized tomography (PET CT) for follow up.

Case Report

A 16 years female presented with a painless mass in her upper neck. The lesion was insidious, progressing gradually for two years. She did not complain of dyspnea, dysphasia or any voice changes. No history of prior irradiation.

Neck examination showed a 2 × 3 cm non-tender mass in the sub-hyoid region at midline, it was soft with smooth surface; the overlying skin was pinchable and normal. The mass was moving with tongue protrusion and deglutition (Figure 1). There were no palpable lymph nodes in the neck. The thyroid gland was not palpable with normal systemic examinations.

The investigations revealed normal thyroid state, ultrasound of thyroid gland showed cystic lesion of 2x3 cm at sub hyoid area, normal thyroid gland and no lymphadenopathy in the neck. A diagnosis of sub hyoid thyroglossal duct cyst was made. The patient underwent Sistrunk's procedure. Intra-operative findings showed a 2 × 3 cm cystic mass in the sub hyoid region, no adhesion to the surrounding structure. (Figure 2 and 3). The patient underwent uneventful recovery and discharged as a day case surgery.

Histopathological study showed typical papillary carcinoma with free resection margins, no angiolymphatic invasion seen.

Positron emission computerized tomography (PET CT) of the neck and whole body was done 6 weeks and one year after surgery, which showed no uptake in thyroid or in the neck neither in any other parts of the body. (Figure 3)

DISCUSSION

The most acceptable theory regarding thyroglossal duct cyst carcinoma origin is that it arises as de novo⁹. Other possible explanations include a multifocal origin in a genetically predisposed individual or a secondary from a papillary carcinoma at thyroid gland¹⁰.

The decision to perform total thyroidectomy in case of thyroglossal duct cyst carcinoma in normally looking thyroid ultrasound with the absence of lymph nodes in the neck was a management dilemma because it leads to life-long thyroid hormone supplement and a chances of hypoparathyroidism; on the other hand, it is needed to make the patient eligible for the radioiodine scans for further diagnosis and radio ablation if needed. It was believed that in the presence of the thyroid gland proper, each ectopic malignant thyroid tissue should be regarded as a metastasis¹¹. However, Renard *et al.*¹² showed that only 6 patients' who underwent total thyroidectomy, revealed carcinoma in the thyroid gland proper out of 43 patients with thyroglossal

cyst carcinoma. Some authors prefer treating thyroglossal duct carcinoma by total thyroidectomy and I 131 ablation^{13,14} while others insist that the malignancy is primary and Sistrunk operation is sufficient followed by long-term observation¹⁵⁻¹⁷.

The clinical presentation of thyroglossal duct cyst carcinoma is usually similar to benign thyroglossal duct cyst and it is often not easy to differentiate between the two by the imaging study. Difficulties arise in the diagnosis of these tumors as they present similarly to benign thyroglossal duct cysts. Most cases are diagnosed postoperatively¹⁸. Furthermore, the sensitivity of fine needle aspiration is about 50–60% in diagnosing carcinoma in such lesion^{19,20}. Shirish S, concluded that fine needle aspiration cytology findings in thyroid duct carcinoma are variable, alone may not be adequate for pre-treatment assessment in all cases²¹. Areej Shahin *et.*²² Found that (61%) cases of surgically resected thyroid duct carcinoma were missed on prior FNA, it showed a diagnostic sensitivity of 62% and a positive predictive value (PPV) of 69% for the diagnosis of thyroid duct carcinoma and it is only moderately sensitive for a preoperative evaluation of thyroid duct carcinoma. Dong Hoon Lee *et al*²³ showed that preoperative FNAC is not routinely necessary for diagnosing thyroid duct carcinoma in children especially given the concerns about possible injury, low sensitivity, and low positive-predictive value. Frozen section examination of the specimen should be considered when malignancy is a consideration. Danilovic *et al.* recommended use of frozen section to diagnose papillary carcinoma due to poor sensitivity of FNAB.

In their study, frozen section correctly diagnosed all cases of thyroglossal duct carcinomas based on suspicious findings on ultrasound²⁴. In our case there was no any suspicious of being malignant, that is why we did not performed frozen section study.

An acceptable guidelines revealed that total thyroidectomy is preferred in high-risk patients based on the following: male patient, more than 45 years in age, history of irradiation, 4 cm mass, extra-capsular invasion, positive nodal state, the presence of cold nodules in the thyroid gland on radioactive scan study²⁵⁻²⁷, in low-risk patients, Sistrunk's operation is enough but requires strict and regular follow up. Patel *et al.*²⁸ found that the 5-year survival rate after Sistrunk's operation for thyroglossal duct cyst carcinoma in a low-risk patient is 100% and there is no need for total thyroidectomy. Furthermore, thyroid suppression therapy after the Sistrunk procedure in low-risk disease has probably no significant benefit⁵.

This case fell into the low-risk category due to less than 45 years of age and less than 4 cm in size with no extra capsular invasion or lymph node involvement. Hence, Sistrunk's operation would be enough. The post-operative Pet CT can be of value in the evaluation of the condition and can be used for follow up.

CONCLUSION

Papillary carcinoma in a thyroglossal duct cyst is rare with controversies in its management. In low-risk patients, a Sistrunk's operation is enough and perfect, but in high-risk patients, more intervention should be done.

Using Pet CT postoperatively is of help in determining the next step.

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Informed consent was taken from the patient and parents in the study.

Disclosure Statement

There is neither financial disclosure nor conflict of interest.



Figure 1. Med neck Cystic lesion at sub hyoid area

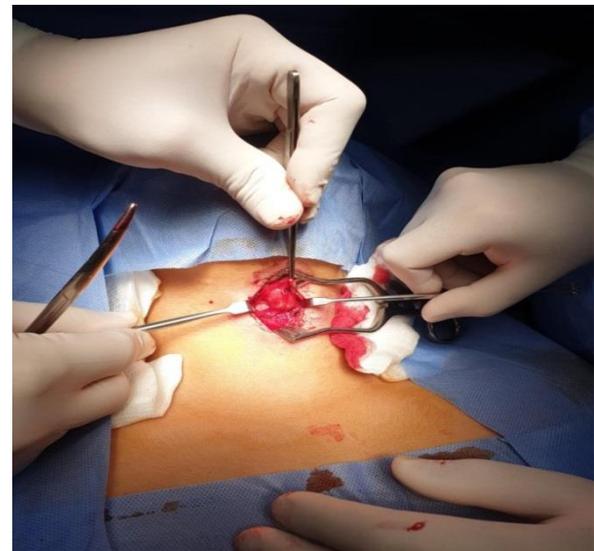


Figure 2,3 surgical procedure



(PET CT) study

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