Case report:

Papillary carcinoma in a recurrent thyroglossal duct cyst

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Abstract

Thyroglossal duct carcinoma (TGDC) is a rare disease with many reported cases. The recurrence in such cases after surgical removal may be seen after cyst excision. Sistrunk's operation is recommended, as it has a very low recurrence rate. The case which present as recurrence demonstrate histological pattern similar to a thyroglossal duct cyst. We present a case of recurrence of thyroglossal cyst, with a solid internal component on ultrasonography (USG). On histopathology the solid internal component proved to be papillary carcinoma. To our knowledge, our paper is the first case of recurrent thyroglossal cyst with a papillary carcinoma preoperatively suspected on USG

Keywords: Thyroglossal duct carcinoma, Papillary carcinoma thyroid (PTC), Ultrasound (USG).

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Introduction

The thyroid gland descends from the foramen cecum to its location at the point below the thyroid cartilage. It leaves behind an epithelial tract known as the thyroglossal tract; the duct is normally obliterated at around the 8th–10th week of gestation, but if the duct fails to involute completely, the remaining epithelial tissue can develop a thyroglossal cyst (TGC). The descent of the thyroid through the anterior midline neck explains several anomalies that relate to thyroid pathology.

Thyroglossal duct cyst is the most common congenital anomaly in thyroid development and occurs in 7% of the adult population (1). Thyroglossal duct cyst accounts for 70% of congenital neck masses(2) and usually manifests as an enlarging painless neck mass in children or young adults. If infected it may present as a red warm painful lump. In most circumstances diagnosis can be made by history and physical examination (3). It may move with swallowing and classically elevates on tongue protrusion. Brentano in 1911 and Uchermann in 1915 are credited as being among the first to describe a neoplasm in a thyroglossal duct remnant; the median age at presentation is 40 years and most patients are asymptomatic (cited by Weiss and Orlich)(4).

The incidence of carcinoma in thyroglossal duct cysts is less than 1%. (5,6) . In a 2004 review of 215 cases, 80% were papillary carcinoma (5,7). Other cancers in decreasing frequency were squamous cell carcinoma, follicular carcinoma, Hürthle cell, insular and rarely anaplastic carcinoma. Medullary

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carcinoma has not been reported in a thyroglossal cyst (5,8-10).

CASE REPORT

A 36 years old lady presented in the out patient department for USG neck. She had undergone a thyroglossal duct cyst excision 12 years back. Now she had again started noticing the swelling in the same region from past 2 years(fig. 1). We performed a detailed USG of neck with a 12 megahertz linear array probe. The USG showed a cystic lesion with a solid internal component and micro calcifications in the solid internal component (fig. 2 a and b). Lateral cervical lymphadenopathy was seen with right level I node. Based on these findings we reported the USG as recurrent thyroglossal cyst with possible PTC. No preoperative FNAC was done in this patient. The patient was operated and the specimen was sent for the histopathological examination. However, initial biopsy report mentioned a thyroglossal cyst. There was no mention of PTC. We requested for revision of the diagnosis, and the reviewing pathologist reported the case as PTC in a thyroglossal cyst! This emphasizes the role of preoperative USG, which can raise the suspicion of malignant change.

Fig. 1. Photograph of the patient showing swelling in the upper neck.

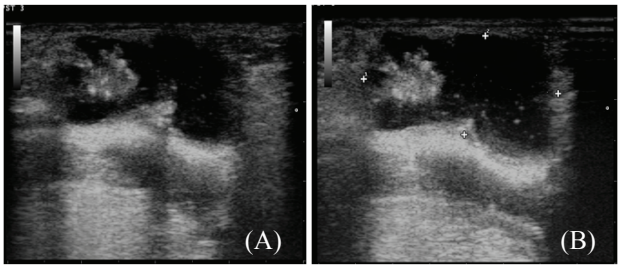

Fig. 2 a and b. A cystic lesion is seen with presence of a small internal solid component. Note the tiny echogenic foci (micro calcifications) in the internal component. Debris is also seen in the fluid of the cyst.

Discussion

Thyroglossal duct cysts develop from persistence of the mid portion of the thyroglossal duct which is an embryonic structure that traces the path of the descent of the thyroid gland. The duct is normally obliterated at around the 8th–10th week of gestation, but if the duct fails to involute completely, the remaining epithelial tissue can develop a thyroglossal cyst (TGC). Previous studies have suggested that this failure to involute occurs in approximately 7% of the population(11).

The etiology of the papillary carcinoma arising in a thyroglossal duct cyst is unclear but, generally, there are two theories which can explain this phenomenon, de novo origin and spread from a primary thyroid gland tumor (12). Most authors support the theory of primary de novo origin by the ectopic thyroid nests of the cyst wall rather than the metastatic spread from a primary thyroid gland tumor through the duct from the thyroid carcinoma. The other theory explains synchronous occurrence of thyroglossal duct cyst carcinoma and thyroid carcinoma, which are reported to be even rarer, as multifocal tumor (13). Papillary carcinoma is the most common malignancy in thyroglossal duct cyst (80%), followed by “mixed” papillary-follicular carcinoma (8%) and squamous cell carcinoma (6 %). The other 6% include very rare cases of epidermoid, Hürthle cell (oxyphillic), follicular, and anaplastic (undifferentiated) carcinomas(14).The concomitant occurrence of papillary carcinoma in the thyroid and thyroglossal cyst have also been reported.

Imaging diagnostic techniques, including ultrasound, scintigraphy and CT, are usually unable to preoperatively diagnose malignant disease(15) and fine needle aspiration yields a correct result in only 66% of the cases (16). The USG in a thyroglossal cyst usually reveals a well defined cystic lesion with anechoic fluid inside. The walls are thin. No internal

vascularity is seen. However, in some cases, the internal fluid may contain debris.

In our case, we observed certain atypical USG imaging features, which arose suspicion of malignancy.These atypical features were the solid internal globular component associated with multiple tiny echogenic foci(microcalcifications) (Fig. 1a and b).

Recently Aculate reported a case of Papillary carcinoma in TGC in a 21 year old woman in whom USG showed a predominantly cystic lesion lying between the hyoid and thyroid cartilages with a central solid component. Post operatively histopathological examination showed a small papillary thyroid carcinoma confined to the wall of the cyst (17). Barton et al reported that carcinoma should be considered in thyroglossal duct cysts that have a mural nodule or calcification on CT (18).

It was concluded that pre-operative USG is a very useful investigation in patients of TGC. Associated thyroid lesions are also evaluated. It can also detect lymphadenopathy with certainty.

Conclusion

Because of the rarity of TGDC, this diagnosis may be missed preoperatively, drastically affecting the appropriateness of the treatment provided. USG can lead to a preoperative diagnosis of thyroglossal duct carcinoma, as a central solid nodular component within the cyst with micro calcifications. USG is also used to guide the FNAC. USG may also be useful tool in post operative follow up. Thus USG should be ordered in the routine preoperative workup of patients with thyroglossal cyst.

This case report with photograph was published with ethical approval.

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