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Papillary Carcinoma in Thyroglossalduct Cyst: an Unusual Case

KEYWORDS: Thyroglossal duct cyst: papillary carcinoma

Dr.V Sudhirbabu	Dr.Sreenivasulu M
Assistant Professor of ENT siddhartha medical college/ GOVT GEN HOSPITAL VIJAYAWADA, A.P.,	Senior Resident SMC, GGH,VIJAYAWADA

ABSTRACT *Thyroglossal ductcyst is commonly encountered in ENT practice. They are treated by Sistrunksoperation. Very rarely Thyroglossal duct cyst may harbor malignancy.in that situations the management protocol is different. We present the case of a 29 year old female with the unexpected finding of a papillary carcinoma arising in a sub hyoid region Thyroglossal duct cyst and is described with special regard to rarity of the localization and the different options in management strategy when carcinoma is found incidentally following surgery.*

1 Introduction

Thyroglossal duct cysts are the most common anomaly in thyroid development. They are twice frequent as branchial cleft abnormalities and, in children, are second only to enlarged cervical lymph nodes as the cause of neck mass. Generally, duct cysts are benign, but 1%of cases may be malignant1.

Most cases of Thyroglossal duct cyst carcinoma are diagnosed during the third and fourth decades of life, and rarely in children less than 14 years of age.2 and 3

Papillary carcinoma is most commonly seen have a favorable prognosis as cervical and distant metastasis is rarely seen. Further management of the patients who are incidentally diagnosed as carcinoma after surgery depends on the spread of tumor.Limited tumors are managed by regular monitoring while the more aggressive ones are managed by total thyroidectomyandradioabalation.4 and 6

2 case report

Baby 29 year old female presented to the ENT OPD of our institute with complaint of painless, midline swelling over the anterior part of upper neck for the last 1year,insidious in onset and gradually progressive in size. There was no history of dysphagia,dyspneoa or hoarseness. There was no history suggestive of hypo or hyper thyroidism. The patient gave no history of radiation exposure. There was no other significant past or personal history. On examination there was a single swelling in the sub hyoid region in the mid line, approximately 3:2 cm in size, oval shape, well defined, and soft in consistency, swelling was non tender, overlying skin was normal in appearance and could be pinched easily. It moved with deglutition and protrusion of tongue. There was no palpable lymphadenopathy. The thyroid gland was clinically normal in shape and size. Rest of systemic examination was normal. A provisional diagnosis of Thyroglossal duct cyst was made. Routine blood investigations and thyroid functions tests (T3, T4, and TSH) were normal.USG neck revealed a 2.7*1.8 cm cystic lesion in the sub hyoid region. Both lobes of thyroid were normal. Aspiration cytology of mass suggestive of thyroglossalcyst. Under general anesthesia, the patient underwent sistrunks operation with removal of the thyroglossal tract and a part of hyoid bone between the lesser cornu along the tract. Complete specimen was sent for histopathologicalexamination. Post-operative period was uneventful. Biopsy report revealed delicate fibroblastic stroma with areas of blood clot and areas show papillary fronds with cells having a hyper chromatic nucleus and pink homogenous colloid like material. Sparse lymphocytes seen in the stroma. Features suggestive of papillary cystadenoma/adenocarcinoma of Thyroid. On 18 th post op day thyroid scan was performed. It did not show any uptake and the thyroid gland was normal. The patient has been a regular follow-up and has not shown any evidence of recurrence.

Thyroidectomy was not performed because there was no history of previous irradiation and carcinoma was limited to the cyst with no cervical metastasis. Post-operative thyroid scan showed normal functioning gland.

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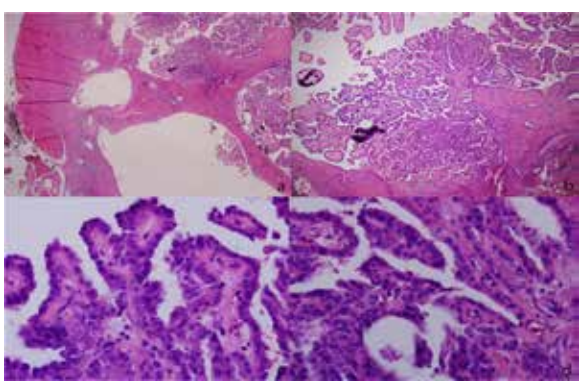

HISTO PATHOLOGICAL SLIDE SHOWING PAPILLARY CARCINOMA OF THYROID

3 Discussion

The thyroid gland descends from the foramen caecum to its location at the point below the thyroid cartilage. It the 5–10th gestational weeks. Incomplete atrophy of the thyroglossal tract or retained epithelial cysts, however, create the basis for the origin of a thyroglossal duct cyst (TGDC).2 and 7 A thyroglossal remnant may be a cyst, a tract or duct, a fistula, or an ectopic thyroid within a cyst or duct leaves behind an epithelial tract known as the thyroglossal tract; this tract usually disappears during. Failure of this tract to close predisposes to the formation of a thyroglossal cyst.3 and 8 60% of TGDC are located between the hyoid bone and the thyroid cartilage, 13% in the substernal region, 24% above the hyoid bone including the sub mental site and 2% are intra-lingual.3, 9 and 10 TGDCs are the most common congenital anomalies in thyroid development, but TGD carcinomas are extremely rare, with 90% of them originating from thyroid remnants.9 and 11 Their cause is unknown and there are no predisposing factors, i.e. neither clinical history nor physical examination can lead to a preoperative diagnosis.12 Papillary types comprise 94%, and less than 5% are of squamous cell origin.6 and 13 Among the various types of neoplasia in TDC, a papillary thyroglossal duct cyst carcinoma has the best favorable prognosis, with occurrence of metastatic lesions in fewer than 2% of cases, while a squamous cell carcinoma has the worst prognosis.3 and 7 The incidence of cervical and distant metastasis is low, with, respectively, a rate of about 8% and 1.3%.11 Generally, there are two theories to explain the thyrogenic origin of TGD adenocarcinomas. Firstly, the de novo theory is based on the fact that in 62% of cases, ectopic thyroid tissue can be identified histopathologically, and this is supported by the absence of a medullary carcinoma in the TGD as it arises from parafollicular cells.8 The second is the metastatic theory which suggests that thyroglossal cyst carcinoma is metastatic from an occult primary thyroid gland, as papillary carcinoma is multifocal in nature.6 Due to these aetiopathogenetic controversies, strict criteria are required to diagnose a primary TDC carcinoma, as proposed by Joseph and Komorowski.5 These are a thyroglossal remnant, ectopic thyroid nests within the cyst wall and a clinically normal thyroid gland.

Regarding the pre-operative diagnosis, finding a TDC carcinoma is unusual. As observed in the present case, patients are generally euthyroid, and the mass is often asymptomatic and not distinguishable from other more common, benign, histological types, in terms of growth, location, size and consistency.3 Regarding the management, some authors believe that well-differentiated incidentally discovered thyroid carcinomas arising in a TDC and limited within their walls, without positive histological margins and cervical metastasis spread, in the presence of a clinically and radiologically normal thyroid gland, can be managed adequately by the Sistrunk's operation, thyroid suppression and strict long-term follow-up.6 and 8 On the other hand, other authors have suggested a more aggressive approach characterized by the Sistrunk procedure, total thyroidectomy, post-operative radioactive iodine therapy and thyroid hormone replacement, based upon the observation that papillary thyroid carcinoma may metastasize through the thyroglossal duct remnant without a lesion being clinically detected in the gland itself.10 and 11

Thyroidectomy is recommended in cases where (a) the thyroid gland is found to be nodular, with a cold nodule in a thyroid iodine uptake scan; (b) enlarged lymph nodes are present, or (c) a history of neck irradiation exists.4

4. Conflict of interest

None.

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