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Papillary Carcinoma of Thyroid in Thyroglossal Cyst with Adenomatoid Changes in both Thyroid lobes – a Rare Presentation – a Case Report

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ABSTRACT

Thyroglossal duct anomalies are the result of a localized persistence of the thyroglossal duct. Though these are the most common congenital anomalies seen in the midline neck, development of carcinoma from a thyroglossal duct cyst is rare and is detected in approximately 1% of cysts.[3] Here we a report a case of a 37 year old female who presented with a midline neck swelling which was diagnosed as a thyroglossal cyst and revealed features of papillary carcinoma of thyroid. Later total thyroidectomy performed in the patient revealed only adenomatoid changes and no focus of carcinoma. The patient was then diagnosed as a case of isolated papillary carcinoma of thyroid in the thyroglossal duct cyst.

I. INTRODUCTION

The thyroid anlage appears in the embryo as a midline structure at the site corresponding to the foramen caecum of the adult tongue. From here, it descends as a component of the thyroglossal duct along the midline to reach its final position in the mid neck. The thyroglossal duct is usually situated anteriorly to the hyoid bone and in the normal course of events it is obliterated and disappears, leaving as a vestige the pyramidal lobe in approximately 40% of normal individuals.[1] Thyroglossal duct anomalies are the result of a localized persistence of the thyroglossal duct. These are the most commonly seen congenital anomalies located in the midline of the neck. However the thyroid tissue present in these anomalies can undergo malignant transformation, usually in the form of papillary carcinoma.[2] The development of carcinoma from a thyroglossal duct cyst is rare and is detected in approximately 1% of cysts.[3] Brentano[4] described the first case of papillary carcinoma in thyroglossal duct cyst in 1911 and since then only about 200 cases of the same have been described in literature.[5] Here we discuss a case of papillary carcinoma seen in thyroglossal duct cyst of a patient who underwent total thyroidectomy to reveal only adenomatoid changes in the gland without any papillary carcinoma focuses.

II. CASE REPORT

A 37 years old female patient presented with midline neck swelling about 3x2 cm. Ultrasonography revealed a thyroglossal cyst with a mass measuring 2x1 cm. Ultrasound guided fine needle cytology suggested a papillary carcinoma of thyroid. Later Sistrunk operation was done and histomorphology confirmed the findings of cytology. After 2 years of that patient was underwent total thyroidectomy. Histology from thyroid gland revealed adenomatoid changes only, no focus of papillary carcinoma was found. Hence it was diagnosed as isolated papillary carcinoma of thyroid in thyroglossal cyst with adenomatoid changes in thyroid gland. The patient recovered fully.[Fig1.1-1.5]

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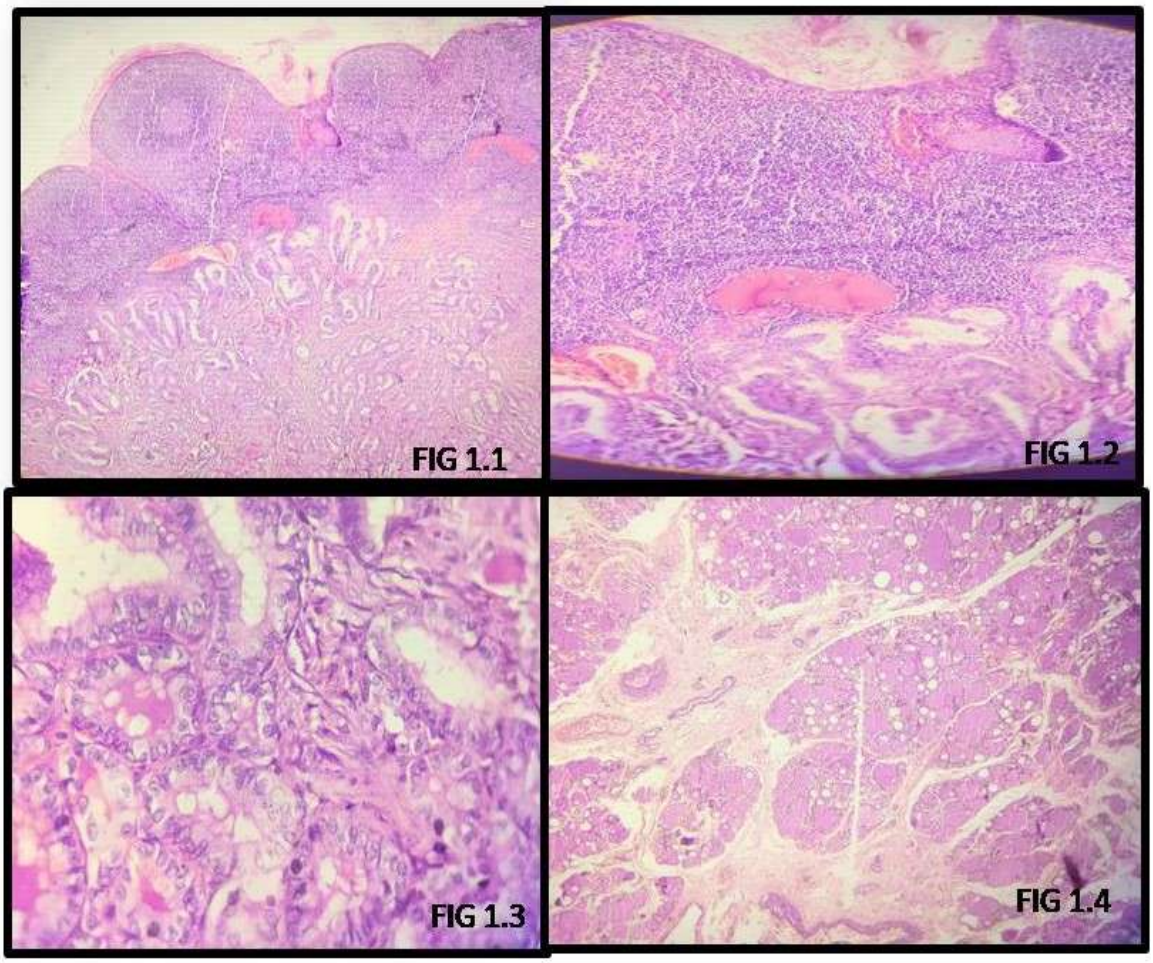

Fig1.1 Cyst wall lined by squamous epithelium with overlying keratin flakes showing invasion by papillary carcinoma thyroid(H&E, x100). Fig 1.2 Cyst wall showing stratified squamous epithelium with invasion by papillary carcinoma(H&E,x100). Fig 1.3 Nuclear features of papillary carcinoma thyroid(H&E,x400). Fig1.4 Adenomatoid follicles lined by simple cuboidal epithelium in both lobes of thyroid in specimen received after total thyroidectomy(H&E, x400).

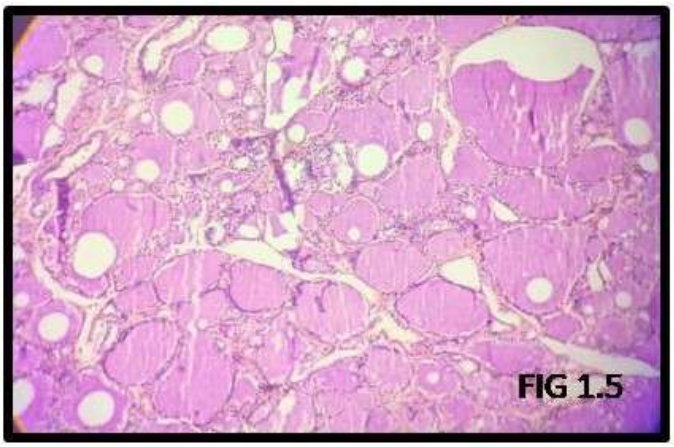

Fig 1.5 Adenomatoid changes as seen in both lobes.(H&E, original magnification x100)

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III. DISCUSSION

Although thyroglossal duct cyst remnants(TGDRs) are the most common congenital midline neck masses, malignant transformation is a rare occurrence and is seen to occur in only about 1 % of the reported cases. Carcinomas in TGDRs tend to have a slight female predilection and the mean age lies in the fourth decade of life.[6] A TGDR is diagnosed on the basis of demonstration of epithelial lining in the cyst/duct and demonstration of normal thyroid follicles in the cyst wall. Therefore, a thyroid carcinoma is generally either of thyroid or squamous cell origin. Thyroid papillary carcinoma is the most common type (80%), followed by mixed papillary/follicular carcinoma (8%) and squamous cell carcinoma (6%).[5]

Widstrom et al established the definitive criteria for diagnosis of a case of primary thyroglossal duct carcinoma: Primary carcinoma of the thyroglossal duct should conform to the following diagnostic criteria: 1. Localization of the carcinoma to a clearly demonstrable thyroglossal cyst or to the anterior midline from the base of the tongue through the hyoid bone to the thyroid isthmus. 2. No carcinoma on histopathological examination of the thyroid gland.[7]

The diagnosis of TGDR carcinomas is usually made postoperatively. Preoperatively on CT and MRI the carcinoma can be seen as a solid nodule in the cyst (the most usual finding), an isolated calcification, an irregular margin, or a thickening in the wall of the cyst. A larger TGDR carcinoma may present as a solid mass in the TGD or as a complex and sometimes invasive lesion in the midline of the neck.[8]

The differentiation between carcinoma arising in ectopic thyroid tissue and a metastatic carcinoma is difficult. The diagnosis can be made indirectly by taking some features into account, such as separate blood supply of the ectopic gland from extra-cervical vessels, no personal history of malignancy, and normal or absent orthotopic thyroid with no history of surgery.[9]

The developmental mechanism of ectopic thyroid malignancies is poorly understood and this poses a challenge in differentiating between isolated malignancies of ectopic thyroid tissue and metastasis from of native thyroid tissue. Various theories exist regarding the origin of these malignancies. Firstly, the de novo theory is based on the fact that in 62% of cases, ectopic thyroid tissue can be identified histo-pathologically, and this is supported by the absence of a medullary carcinoma in the TGD as it arises from parafollicular cells.[10] Papillary carcinomas that develop in thyroglossal duct cysts and synchronously in the thyroid gland can be explained by the fact that papillary carcinomas can synchronously develop as multifocal. Alternatively, some authors evaluate them as a metastasis from the primary focus in the thyroid gland into a thyroglossal duct cyst.[11]

Reported treatment options for cancers arising in TGDR carcinomas have included tumorectomy, Sistrunk's procedure, or Sistrunk's procedure associated with total (or subtotal) thyroidectomy. Studies in literature do not provide sufficient information on the frequency of the development of carcinoma in the thyroid gland and thyroglossal duct cyst and their relationship with each other because they generally include a single or a few patients with papillary thyroid carcinoma and because of the rarity of such cases. For these reasons the application of total thyroidectomy to the management of TGDR carcinomas is still controversial. Ramirez Plaza et al. proposed an algorithm for treatment of papillary carcinoma in TGDR, with a simple Sistrunk procedure for patients less than 45 years of age with tumors less than 1.5 cm confined to the cyst and an ultrasonographically normal thyroid gland with no suspicious lymph nodes. A total thyroidectomy (with compartment-oriented neck dissection only if lymph node metastases are found on ultrasound or during surgery) followed by radioiodine is recommended for those not fitting these criteria.[12] One advantage of performing a total thyroidectomy is to allow follow-up with precise thyroglobulin measurements. The risks of surgery and the need for long-term treatment by thyroxine may outweigh this advantage in thyroid microcarcinoma, however, because of the low risk of recurrence for these tumors. Prophylactic neck dissection is controversial.[13]

IV. CONCLUSION

In conclusion we report a rare case of isolated primary papillary carcinoma of the thyroglossal duct cyst without any malignancy in the native thyroid tissue. Sistrunk operation was done for thyroglossal cyst tumour and after that total thyroidectomy was also done for that patient. The tumour in thyroglossal cyst was diagnosed as papillary carcinoma of thyroid but in thyroid it was found only adenomatoid changes without any tumour focus. Hence it was a case of isolated papillary carcinoma of thyroid in thyroglossal cyst.

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