PAPILLARY CARCINOMA THYROID IN THYROGLOSSAL CYST- A CASE REPORT

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INTRODUCTION

Thyroglossal cyst is a congenital anomaly formed as a result of persistence of thyroglossal duct during development. The development of malignancy in a thyroglossal cyst is an extremely rare condition with an incidence of 1% of all the cases of thyroglossal cyst.[1,2] This article shows a rare case of occurrence of papillary carcinoma thyroid in a case of thyroglossal cyst.

PRESENTATION OF CASE

A 28-year-old female patient came with chief complaint of single midline painless cystic swelling in front of the neck which started 6 years back, insidious in onset, gradually progressive, started as a small swelling and reached present size. There were no signs and symptoms suggestive of hyperthyroidism or hypothyroidism. There were no pressure symptoms like dyspnoea, dysphagia, and hoarseness of voice (presence of which indicates malignancy).[3] There was no history of loss of weight and loss of appetite associated with the swelling.

On inspection, a single midline ovoid swelling of size 8*4 cms was noted in the anterior aspect of neck which moved with deglutition and protrusion of tongue. No visible pulsations, no engorgement of veins, no scars and no sinuses were noted. On palpation, a single midline painless swelling of size 8.5*6 cms was present in front of the neck, which is cystic in consistency, with nodular surface, freely mobile, moving up with deglutition and protrusion of tongue. No regional cervical lymph nodes were palpable. (Figure 1).

Thyroid function tests and other laboratory values were normal. Ultrasonography of the neck showed multiple unechoic midline cystic lesions in front of the neck of 2.5 cm*1.5 cm dimension in the subcutaneous plane, suggestive of thyroglossal cyst. Bilateral thyroid glands were normal. In order to further evaluate and plan the appropriate surgical procedure, MRI of the neck was done. MRI showed an elongated, well circumscribed lobulated midline cystic lesion extending from hyoid up to thyroid with lesion splaying the strap muscles and abutting the sternocleidomastoid laterally, with features suggestive of thyroglossal dust cyst. FNAC from the swelling showed features suggestive of thyroglossal cyst or cystic nodular goiter. (Figure 2)

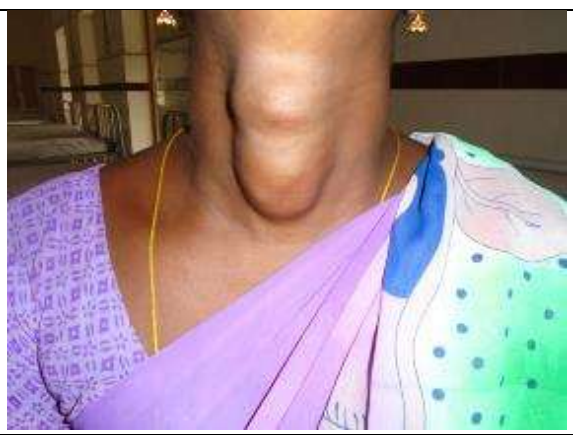

Figure 1. A Well-Defined Painless Midline Cystic Lesion in Front of the Neck

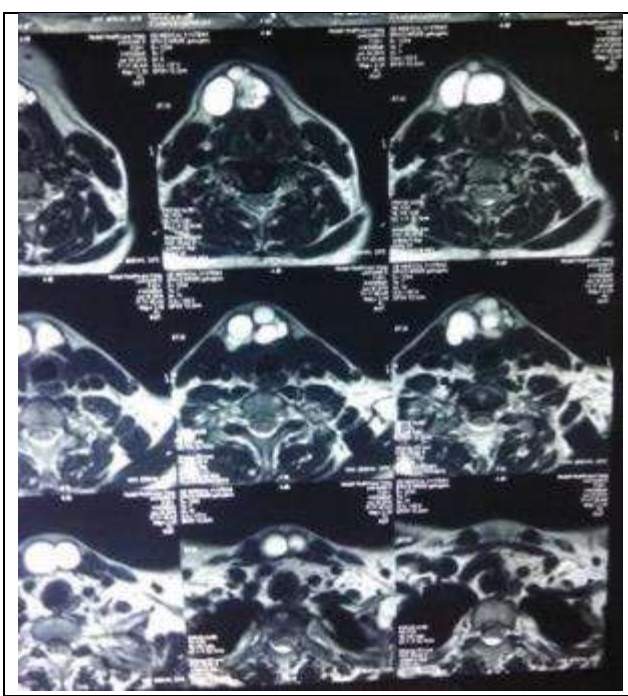

Figure 2. MRI Neck Showing Anterior Midline Cystic Lesion with Features Suggestive of Thyroglossal Cyst

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DIFFERENTIAL DIAGNOSIS

1. Nodular goiter.
2. Solitary goiter arising from isthmus of thyroid.
3. An enlarged lymph node.
4. Dermoid cyst.

DISCUSSION OF MANAGEMENT

The patient was then posted for surgery; Sistrunk procedure was done. A mass of 5*2.5*1 cms was excised including the entire duct from the thyroid gland up to the level of foramen caecum along with the middle portion of hyoid bone.[5] The association of recurrence of thyroglossal duct cysts without removal of hyoid bone central portion has been well documented.[6,7]

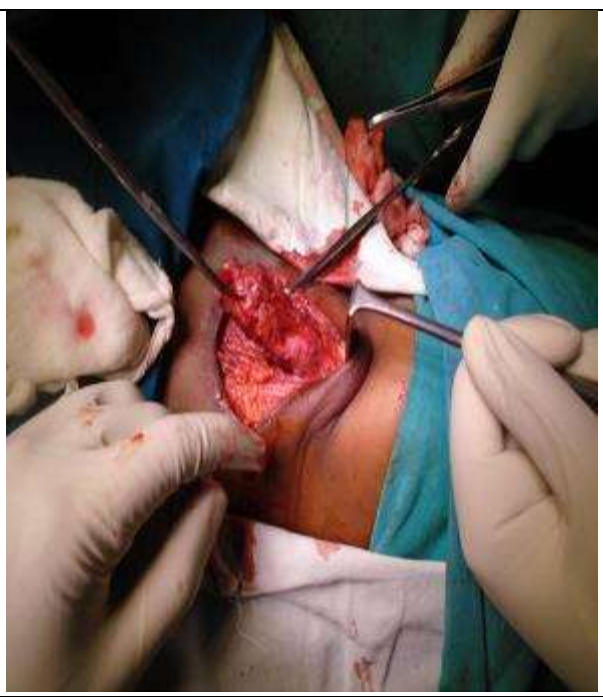

Figure 3. Intra-Op Findings of Sistrunk Procedure

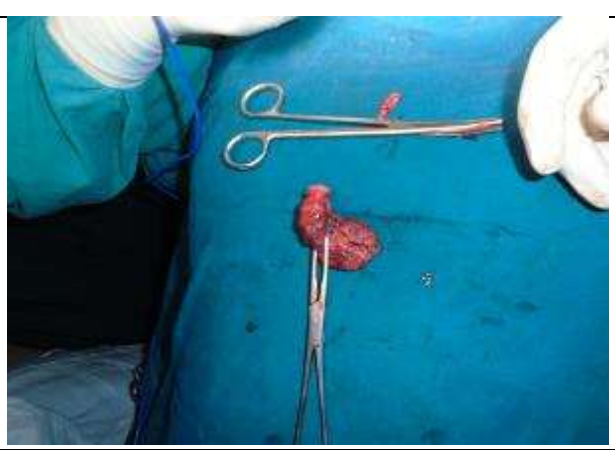

Figure 4. Thyroglossal Duct Cyst Following Excision

PATHOLOGICAL DISCUSSION

Histopathological examination showed thyroglossal cyst lined by cuboidal epithelium with papillary formations and follicles. Some of the cells showed ground glass nuclei with nuclear inclusions and grooves. Thus, the features were suggestive of Papillary Carcinoma thyroid in Thyroglossal Duct Cyst. Microscopic examination of the specimen in this case revealed infiltration of the cystic wall and normal thyroid gland, confirming the diagnosis of primary papillary carcinoma arising in a thyroglossal duct cyst.

Thyroid gland is the first endocrine gland to develop, which arises embryologically from the floor of the pharyngeal gut (foramen caecum)[8] and descends as a bilobed diverticulum remaining connected to the gut by a narrow canal, the thyroglossal duct. Normally, the thyroglossal duct obliterates and disappears by the tenth week of gestation. If it fails to involute, it may remain in the form a cyst, a tract or duct or as ectopic thyroid within a cyst or a duct which is localized in the midline between base of the tongue and the pyramidal lobe of the thyroid gland.[9] A remnant of the thyroglossal duct, usually a cyst is the most common congenital anomaly of the thyroid gland development.[10] The percentage of ectopic thyroid tissue within thyroglossal duct cyst usually ranges from 1.5% to 45% cases.[1] Clinically, most thyroglossal duct cysts are benign and as midline, painless, slow-growing neck masses.[10] It is usually seen in children and adolescents and only one-third of them are at a minimal age of 20. Criteria for the diagnosis of thyroglossal cyst are that it must be located in the median region of the neck and be composed cuboidal cells; both lymphatic tissue and normal thyroid follicles should be present in the cystic wall. These are considered to be developing as a result of mutations caused in genes such as the thyroid transcription factors T1TF1, T1TF2 and PAX8 responsible for the development of thyroid follicular cells.[11] Common complications of thyroglossal cyst are infection and sinus formation, but the development of malignancy in the cyst is a rare case.[12] Thyroid carcinoma occurring in a thyroglossal cyst is a rare entity with only few cases reported in the literature, with papillary carcinoma thyroid being the most common type,[13] followed by mixed papillary-follicular type and less than 5% being squamous cell type.[14,15] The predominant theories described for the development of this malignancy are not clear, but it could either be a metastatic spread from occult primary or spontaneous development from ectopic thyroid tissue present within thyroglossal cyst.[16] Malignancy occurs only in 1% of thyroglossal duct cysts and occurs most commonly in women,[17] with the mean age being during the fourth decade of life.[18] The rate of co-existence of thyroglossal duct cyst and thyroid cancer is between 0-25%.[19]

According to the Widström et al. criteria, the primary carcinoma of thyroglossal duct cyst is diagnosed by the histopathological confirmation of thyroglossal duct carcinoma, normal epithelial lining of the thyroglossal duct, normal thyroid follicles within the walls of the cyst, presence of normal thyroid tissue adjacent to tumour, and no abnormalities in the thyroid gland, most of which are present in this case.[21]

The primary management of the thyroglossal duct carcinoma is still controversial, regarding the management of thyroid gland along with thyroglossal gland.[20] While some authors consider only Sistrunk procedure as the

primary management but others consider including total or near-total thyroidectomy in the management of thyroglossal duct carcinoma.[22] The rationale for thyroidectomy to be included in the thyroglossal cyst carcinoma is based on: presence of occult primary in thyroid gland proper, use of radioactive iodine as an adjuvant treatment, and role of thyroglobulin as a serum marker for post-operative follow-up.[8] However, Renard et al. demonstrated that out of 43 patients with thyroglossal cyst carcinoma who underwent total thyroidectomy only 6 patients showed thyroid carcinoma in thyroid gland proper.[23]

Some literature states that surgical approach in these patients should be based on risk group stratification. Accordingly, Sistrunk procedure is considered as the only procedure in the setting of a clinically and radiologically normal thyroid gland in low-risk patients with low-risk tumours, for patients who are age <45 years, with no history of radiation exposure, a tumour size <4.0 cm, no soft tissue invasion, no lymphadenopathy, and the absence of aggressive tumour histology.[24] Tharmabala and colleagues proposed a risk stratification groups into three classifications: for low-risk, observe and wait; for moderate risk, total thyroidectomy, hormonal suppressing treatment, and radioactive iodine; for the high-risk group, vertical lymph node dissection in addition to the other treatments.[21]

Metastatic spread is uncommon, and the prognosis is usually good.[15,26] Regional lymph node metastasis of thyroglossal duct carcinoma occurs only in 7% cases and local spread is usually rare.[9] Careful long term follow-up of papillary carcinoma of thyroglossal duct carcinoma is very important as it is a low grade malignancy and recurrences can be treated adequately with adequate follow-up. All the patients must have a six-month follow-up in the first year and there after annually.[27] Follow-up should include thorough physical examination, ultrasound of the surgical site and thyroid gland and thyroid scan.[28]

DISCUSSION OF MANAGEMENT

In this case, due to the low-risk risk stratification group, age <45 years, no prior radiation exposure, no cervical lymph node involvement, absence of aggressive tumour histology, we considered only Sistrunk procedure as the primary management with regular follow-up, thyroglobulin levels were used as a regular follow up marker and radioactive iodine was given post-operatively.[25] Post operatively patient advised radioactive iodine treatment with regular follow-up for every six months in the first year and annually thereafter. Thyroglobulin levels were done post-operatively and were observed to be within the normal change.

FINAL DIAGNOSIS

Papillary Carcinoma Thyroid in Thyroglossal Cyst.

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