

# Papillary Carcinoma of Lateral Thyroglossal Duct Cyst With De Novo Papillary Carcinoma of Thyroid: A Rare Case Report

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## Abstract

Thyroglossal duct cysts are commonly seen in the midline of the neck and have been reported to be present in 7% of those who have midline swelling. Carcinoma in the thyroglossal duct cyst is extremely uncommon, occurring in fewer than 1% of cases. There have been relatively few examples of it developing in a lateral site recorded in the literature. This case represents a 25-year-old girl with papillary carcinoma of left lateral thyroglossal duct cyst with De Novo papillary carcinoma of right lobe of thyroid.

**Keywords:** Lateral Thyroglossal Duct Cyst; Midline Swelling; Papillary Carcinoma of Thyroid.

## Introduction

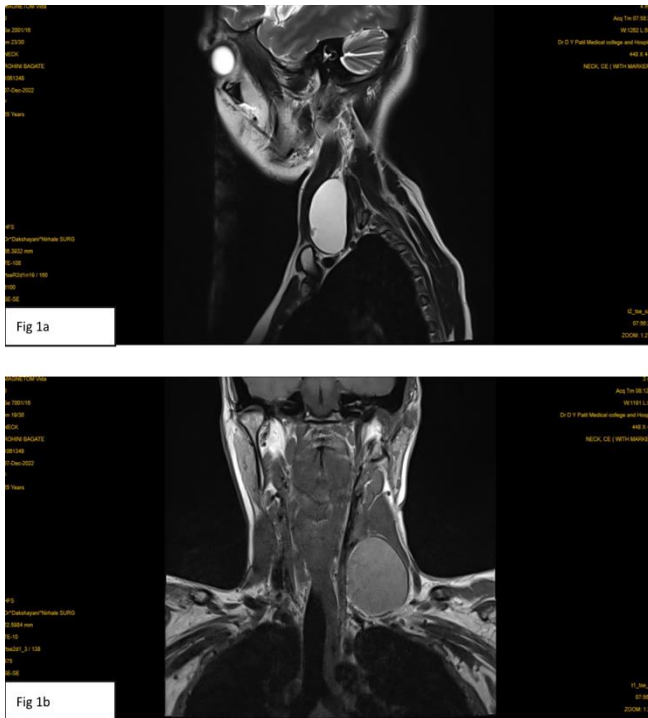
The thyroid gland develops as a result of endodermal cells invading the midline during the third and fourth weeks of embryological development. From the base of the tongue to the front of the neck, in the direction of the first and second tracheal rings, this epithelial invagination descends along the midline.<sup>1</sup> The hyoid bone forms in the second and third arch's mesoderm, allowing the thyroglossal tract to pass ventral to it. Between the eighth and tenth week of pregnancy, the thyroglossal duct obliterates its course. The thyroglossal cyst, which is thought to be present in 7% of people with midline swelling, is caused by the inability to completely obliterate portions of this canal.<sup>2</sup>

Thyroglossal duct cysts usually appear in the neck as an asymptomatic midline swelling. Though there are relatively few occurrences of it occurring in a lateral site that have been documented.<sup>3</sup> Less than 1% of them have concurrent thyroid cancer that develops in an untreated thyroglossal duct cyst.<sup>4</sup> Signs of cancer include dysphagia, dysphonia, and gradual weight loss with an abrupt increase in the size of the tumour. Most patients are asymptomatic, with a median age at presentation of 40 years.<sup>5</sup> The percentage of false negatives in fine needle aspiration cytology is significant. During the histological examination of the resected cyst, primary papillary cancer in the thyroglossal duct cyst was discovered by accident. Because this form of carcinoma is so uncommon, there are no set standards for a typical treatment plan.<sup>3</sup>

## Case Report

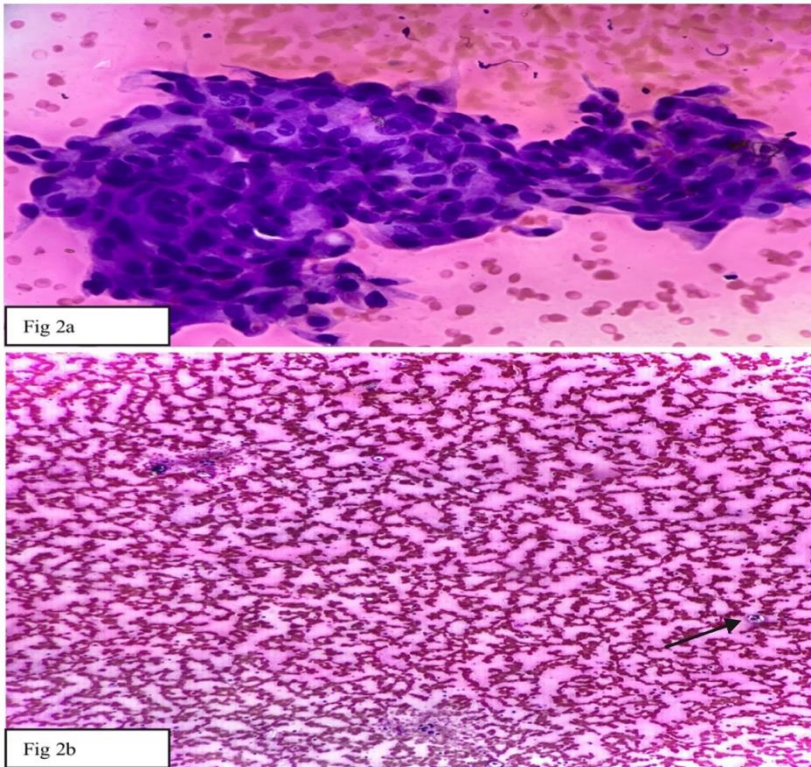
A 25-year-old female patient complained of swelling in the left lateral area of her neck and pain during eating, that had been present for three months but now swelling was rapidly increasing in size. Upon inspection, the enlargement in the left lateral portion of the neck measured 4 to 5 cm which is associated with pain during swallowing and slightly moves with protrusion of tongue. Clinically, there was no enlargement of the thyroid or any signs and symptoms of hyper or hypothyroidism. No palpable cervical lymph nodes present. Her menstrual history has been regular, with no significant previous, familial, or personal history. The patient's routine blood workup and thyroid function tests were both within normal limits. The thyroid lobes and isthmus appears normal, but an ultrasound of the neck revealed a cystic lesion measuring size 46×23×18 MM in the anterior lateral part of the neck. Both lobes of thyroid and isthmus appearing normal in size, shape, echotexture and vascularity.

Radiological diagnosis of brachial cyst was given and histopathology and cytology correlation was advised for confirmation. Further on Magnetic resonance imaging, which was inveterate the same [Figure 1].



**Figure 1:** a) Magnetic resonance imaging showing a heterogeneous swelling in the neck. b) Magnetic resonance imaging showing a heterogeneous swelling present in left lateral part of neck.

The results of the fine needle aspiration cytology revealed mostly haemorrhagic and proteinaceous background with a few mucinous flakes, few tiny clusters of benign epithelial cells and cyst macrophages suggested the diagnosis of a benign cystic lesion with histopathology correlation.[Figure 2 a,b] After being excised, this cyst was sent for a histopathological analysis. The gross cyst measured 4x3x2 cm in diameter. When the gross cyst was cut 5 ml of haemorrhagic fluid oozed out and revealed grey-white cyst wall and papillary excrescences inside the cyst wall picture. [Figure 3] The submitted cyst wall sections was examined which reveal columnar epithelial cells lining of the cyst as well as few dilated thyroid follicles filled with colloid eosinophilic material. [Figure 4a] The section further shows focus of papillary projections that are lined by malignant cells which are having eosinophilic cytoplasm, overlapping nuclei, nuclear grooves and pale chromatin.[Figure 4 b,c] Consequently, the patient was scheduled for a complete thyroidectomy with radical neck dissection surgery when the diagnosis of primary papillary carcinoma in lateral thyroglossal duct cyst was made. Total thyroidectomy specimen along with radical neck dissection was then sent for histopathological examination. Thyroid gland measuring 4 x 3 x 1.5cm, externally capsulated and on cut section showed two tiny whitish areas in right lobe. Thyroid gland was serially sectioned and submitted in toto. Microscopy shows thyroid tissue and a small focus of papillary carcinoma in the thyroid stroma. [Figure 4d] Total 18 lymph nodes were dissected and all were free of tumor on microscopy. The patient was discharged postoperatively and is advised followup.

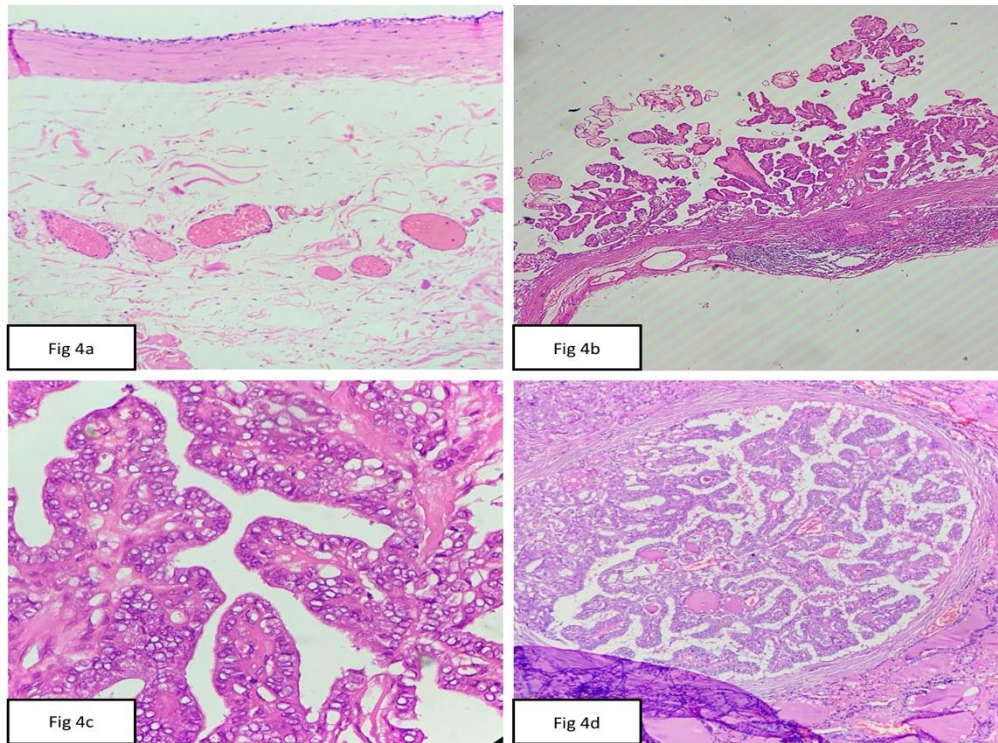


**Figure 2:** a) Fine needle aspiration cytology showing tiny clusters of benign epithelial cells. b) Fine needle aspiration cytology showing tiny clusters along with cyst macrophage (black arrow)



**Fig 3**

**Figure 3:** Gross image showing papillary excrescences inside the cyst wall.



**Figure 4:** a) Section from cyst wall shows benign epithelial lining. b) Section from cyst wall showing papillary projections that are lined by malignant cells.(4x) c) High power view showing classical papillary carcinoma nuclear features. d) Section from thyroid showing small focus of papillary carcinoma in the thyroid.

Consent of the patient was taken for the purpose of study and publication.

## Discussion

The majority of patients with papillary thyroid carcinoma of the thyroglossal duct cyst are asymptomatic and typically appear with dysphagia, dysphonia, increasing weight loss and a rapidly growing mass. The median age at presentation is 40 years.<sup>5</sup> Our is a 25-year-old woman who was presented with dysphagia as a result of swelling in the lateral part of her neck since three months, whose size was increasing rapidly and was slightly movable with protrusion of tongue.

Allard studied 381 cases of midline TGDCs and found out that majority of them were infrahyoid comprising 60.9%, suprahyoid in 24.1%, suprasternal in 12.9% and lingual in 2.1% of them. Rarely, TGDCs may manifest in a non-classical manner. Thabet studied 22 patients of thyroglossal duct cyst to look for atypical presentation. In his study he found that 6 cases showed variable presentation i.e one each presented as thyroglossal duct cyst with intralaryngeal extension, intralingual thyroglossal duct cyst, thyroglossal duct cyst within anterior neck compartment, thyroglossal duct cyst with intracystic solid mass, inferiorly located thyroglossal duct cyst, laterally located thyroglossal duct cyst.<sup>7</sup> In our case the swelling was present in the infrahyoid and in left lateral aspect of neck. Few reports in the literature described laterally located TGDCs along the anterior sternocleidomastoid muscle border and included TGDC in the differential diagnosis of branchial cleft cysts.<sup>7</sup> In our case the swelling was present deep to the left sternocleidomastoid muscle making it challenging to diagnose thyroglossal duct cyst radiologically. However it is difficult to differentiate thyroglossal duct cyst from other cysts like brachial cysts, lymphangiomas, epidermoid cysts, dermoid cysts and hydatid cysts clinically and radiologically.<sup>8</sup>

Thyroglossal duct cyst in the lateral aspect of the neck is a rare finding and thyroglossal duct cyst which develop primary carcinomas within are extremely rare, which occurs in less than 1% of thyroglossal duct cyst which was seen in our case.<sup>3</sup> Their cause is unknown and with no predisposing factors i.e neither clinical history, nor physical examination can lead to pre-operative diagnosis.<sup>9</sup>

The cytological examination obtained by fine needle aspiration is only diagnostic in 66% of the cases.<sup>10</sup> It presents same as that of benign cyst which represents the most frequent benign congenital lesion of the neck.

The pathophysiology of thyroglossal duct cyst carcinomas has been the subject of two theories first is direct metastasis of a papillary thyroid carcinoma and second one is Carcinoma De Novo from ectopic thyroid tissue in the cyst. In our case, the thyroid gland further displayed a tiny foci of papillary carcinoma invading the thyroid stroma, making its etiology undistinguishable.<sup>9</sup> In our case its origin is most probably De Novo as the thyroglossal duct cyst is present in the lateral side having papillary carcinoma along with benign dilated thyroid follicles and the foci of papillary thyroid carcinoma which we found in thyroid gland is present in the right lobe of thyroid along with no lymph node involvement.

Though many study suggest to perform total thyroidectomy in all cases of primary papillary carcinoma in thyroglossal duct cyst.<sup>3</sup> The definitive management of thyroglossal duct cyst carcinomas remains uncertain due to rarity of lesion with concerns regarding the treatment strategy for management of the thyroid gland.

The atypical presentation of thyroglossal duct cyst is very rare finding, in our case the papillary carcinoma of thyroglossal duct cyst was present in left lateral side of neck along with De Novo papillary thyroid carcinoma of right lobe of thyroid. There are very few case reported till date in literature.

## Conclusion

Atypical presentation of thyroglossal duct cyst is very rare finding, in our case the papillary carcinoma of thyroglossal duct cyst was present in left lateral side of neck along with De Novo papillary thyroid carcinoma of right lobe of thyroid. It is diagnosed post operatively as an incidental finding on histopathological examination. The management of these cases needs a multidisciplinary medical team for proper treatment approach.

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