



Case Report

**Nonfunctional Cystic Parathyroid Lesion and Papillary Carcinoma
Thyroid - A Rare Co-Occurrence**

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ABSTRACT

*Cystic swelling of the parathyroid glands is a rare pathology of the spectrum of parathyroid gland diseases. It accounts for around 0.4% of various parathyroid pathologies. Here we report a case of papillary carcinoma thyroid and cystic parathyroid lesion, of which the pre-operative cytology of the cystic lesion was misleading. A 52 years old female presented with complaints of progressive swelling in the left anterior neck for four years and difficulty swallowing solids over the past three months. No other associated symptoms were noted. A High-resolution ultrasound revealed multiple hypoechoic nodules with micro-calcifications in the right lobe thyroid largest measuring 1.5*1 cm, TIRADS V, and another anechoic lesion adjacent to the left lobe of the thyroid. In addition, USG-guided FNAC suggested a Bethesda V lesion in the right lobe, and FNAC from the left lobe and cystic lesion were suggestive of colloid goiter. The patient underwent a Total thyroidectomy with excision of the cyst. Final histopathology showed multifocal papillary carcinoma thyroid, a follicular variant with the largest foci of 1 cm; the cyst showed features of clear cells of parathyroid origin. Coexistence of papillary is rarely reported in the literature, and it could be merely a coincidence. There should be a high clinical suspicion of cystic parathyroid lesions in case of atypical findings on sonography or contrast-enhanced computed tomography, and further substantiated with cyst fluid cytology and PTH levels will further guide to the diagnosis of cystic parathyroid lesions in most of the cases.*

Introduction

Cystic swelling of the parathyroid glands is a rare pathology of the spectrum of parathyroid gland diseases. It accounts for around 0.4% of various parathyroid pathologies (1). So far, around 360 cases of cystic parathyroid glands have been reported in the literature. Two types of clinical entities exist in this kind of pathology, functional or nonfunctional, of which the latter is the more common clinical entity. Patients with cystic parathyroid lesions have varied clinical presentations or symptoms. There are no specific diagnostic features; hence the pre-operative diagnosis of parathyroid cysts is often challenging (2). Here we report a

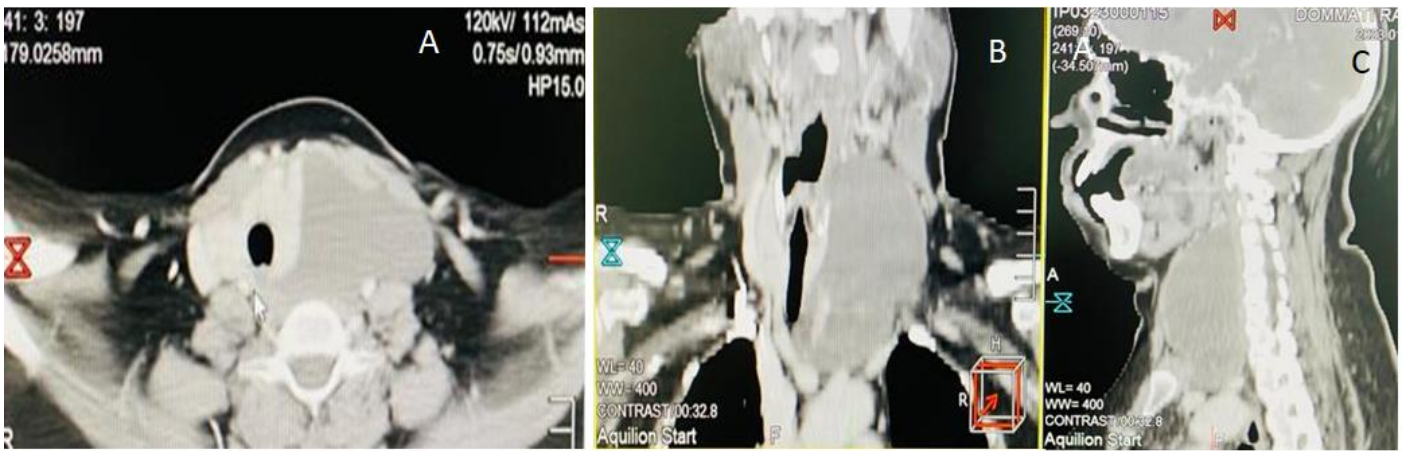
rare case of papillary carcinoma thyroid and cystic parathyroid lesion, of which the pre-operative cytology of the cystic lesion was misleading.

Case capsule

A 52 years old female presented with complaints of progressive swelling in the left anterior neck for four years and difficulty swallowing solids over the past three months. No other associated symptoms were noted. Otherwise, her medical and surgical history was not contributory. The patient underwent a detailed ENT examination; a single ill-defined cystic swelling in the left anterior neck extended from the angle of the mandible above to the clavicle below, laterally extending beyond the posterior border of the left sternocleidomastoid muscle. In addition, multiple small nodules were palpable in the right lobe of the thyroid. The patient was clinically and biochemically euthyroid and normocalcemic (8.6mg/dl).

A High-resolution ultrasound revealed multiple hypoechoic nodules with micro-calcifications in the right lobe thyroid largest measuring 1.5*1 cm, TIRADS V, and another anechoic lesion adjacent to the left lobe of the thyroid. In addition, USG-guided FNAC suggested a Bethesda V lesion in the right lobe, and FNAC from the left lobe and cystic lesion were suggestive of colloid goitre. Because of the upper mediastinal extension of the lesion, a contrast-enhanced computed tomography of the neck and thorax was done, which was suggestive of a cystic mass measuring 10 × 7 × 8.5 cm adjacent to the left lobe thyroid displacing the left internal jugular vein and left common carotid. Along with nodular opacity and microcalcification in the right lobe thyroid. The superior and inferior extent from C3 cervical vertebrae to the aortic arch (Figure 1). Flexible laryngoscopy revealed no laryngeal or upper tracheal pathology except for the gross deviation of the laryngeal framework towards the right.

The patient underwent a Total thyroidectomy with excision of the cyst. Intra and immediate postoperative period was uneventful. The patient was discharged on postoperative day five with no signs of hypocalcemia. Final histopathology showed multifocal papillary carcinoma thyroid, a follicular variant with the largest foci of 1 cm; the cyst showed features of clear cells and a parathyroid gland along the wall. (Figure 2)

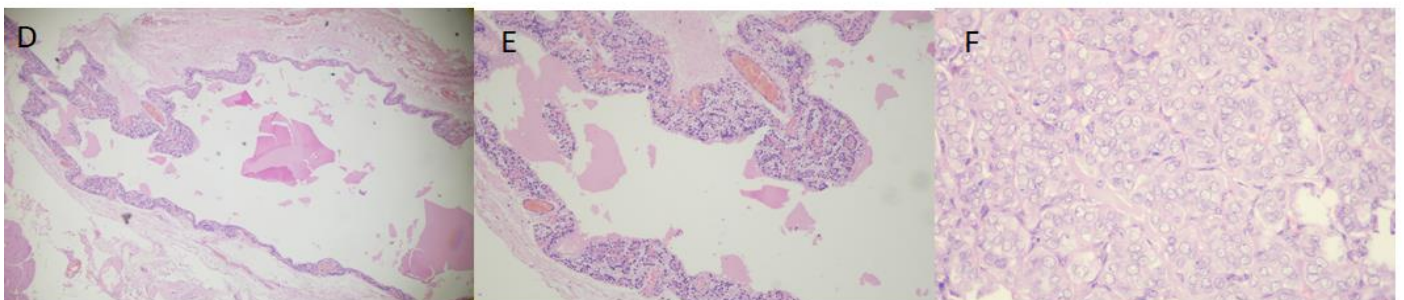


A. CT neck demonstrating left cystic mass measuring 10 × 7 × 8.5 cm adjacent to left lobe thyroid displacing left internal jugular vein and left common. Along with nodular opacity in the right lobe thyroid.
 B,C. CT neck showing the superior and inferior extent from C3 cervical vertebrae and inferior up to the aortic arch.

Figure 1: Depicts the Contrast enhanced computer tomography of the lesion



A. Exposure of the cyst by separating the strap muscles, B. Excision of cyst after identifying the Left RLN (Instrument pointing) along its course. C. Total thyroidectomy specimen.



D. 40x magnification representative image showing cyst wall lined by clear cells with parathyroid gland in the wall without atypia or mitosis, E. 100X magnification representative image showing cyst wall lined by clear cells with parathyroid gland in the wall without atypia or mitosis F. Sections shows foci of papillary carcinoma thyroid (Follicular variant)

Figure 2: Depicts the surgical excision and histopathology of the lesion.

Discussion

Cystic parathyroid lesions represent around 0.5 - 1% of the spectrum of parathyroid diseases. A systematic review and meta-analysis by Papavramidis et al. report 359 documented cystic parathyroid lesions in the literature, and 61% are nonfunctional cysts (2). A single institutional 20-year review from France reported an incidence of 1.3% of parathyroid pathologies. Typically, small cystic parathyroid lesions are incidental findings during surgery, and large cystic lesions present as palpable neck masses, most likely diagnosed on final histopathology (3). Another study from France reported the incidence of parathyroid cysts as 1% of parathyroid pathologies (4).

There is still no consensus on the pathophysiology of the cystic lesions of the parathyroid. They are thought to arise due to central degeneration of the parathyroid adenoma or coalescence of the micro cysts (5). However, another theory is that they are likely of branchial origin (3).

Clinically present as lateral neck swellings anywhere from the hyoid to the upper mediastinum. Lesions can be functional and present as primary hyperparathyroidism, which accounts for a significantly lower percentage of cases. Others may present with compressive symptoms of the size of the lesion. Very few cases have been reported to present a change in voice associated with vocal cord palsy (6).

Imaging alone cannot distinguish cystic parathyroid lesions in otherwise normo-calcemic patients. Sonography may show an echogenic plane separating the lesion from the thyroid gland, and there may be an evident feeding vessel on the Doppler (7). Cystic parathyroid adenomas adjacent to the thyroid may be frequently misinterpreted as cystic thyroid lesions, which was the scenario in our case. In addition, there may be no or minimal uptake of MIBI in cystic parathyroid lesions, which may further add to the confusion (8). Fluid aspirated from the cyst and PTH levels may explain the parathyroid origin. However, although the fluid is high in PTH, the cystic lesion may not be functional (i.e., secreting high parathyroid hormone levels) (2).

Excision of the cyst is the gold standard treatment option and also for the correct histopathology diagnosis of the cyst. Other less popular treatment options are aspiration of the cyst or injection of the sclerosing agent. In addition, intraoperative PTH monitoring is suggested in the case of functional parathyroid cysts to explore the second adenoma, as most parathyroid cysts are nonfunctional.

In our case, the patient has a Bethesda V lesion of the right lobe of the thyroid along with a cystic lesion of the left parathyroid. The coexistence of these two conditions is rarely reported in the literature, and it could be merely a coincidence.

There should be a high clinical suspicion of cystic parathyroid lesions in case of atypical findings on sonography or contrast-enhanced computed tomography, and further substantiated with cyst fluid cytology and PTH levels will further guide to the diagnosis of cystic parathyroid lesions in most of the cases.

Key Points

- Cytological diagnosis of cystic lesions in the neck can be deceptive. Cystic parathyroid lesions may be reported as thyroid lesions.
- Atypical USG and CT findings of cystic lesions in relation to the thyroid gland should raise suspicion of cystic parathyroid lesions.
- Cystic fluid analysis for PTH levels may give a better pre-operative diagnosis of cystic parathyroid adenomas.
- MIBI SPECT CT may not be helpful in nonfunctional cystic parathyroid adenomas.

Conflict of Interest

Authors declare no conflict of interest

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