CASE REPORT

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CERVICAL CYST – A TALE OF THE UNEXPECTED

Y. Vlahov[1], E. Mollova[1], K. Vidinov[2], G. Mateva[3], M. Mladenov[2], P. Bochev[3], A. M. Borissova[1]

[1]Clinic of Endocrinology and Metabolic Diseases, University Hospital Sofiamed – Sofia, Bulgaria
[2]Department of General and Thoracic Surgery, University Hospital Sofiamed – Sofia, Bulgaria
[3]Department of Nuclear Medicine, University Hospital Acibadem City Clinic Mladost – Sofia, Bulgaria

Abstract. *Primary hyperparathyroidism (PHPT) is the third most common endocrine disease after diabetes and osteoporosis. It is more common in women and its prevalence increases with age. PHPT is associated with parathyroid adenoma or cancer or paraneoplastic parathyroid hormone (PTH) production. PHPT can be asymptomatic or present with nephrolithiasis, peptic ulcers, pancreatitis, bone lesions and rarely – as a palpable formation in the cervical area. We present a 73-year-old female patient with cystic cervical lesion with high levels of PTH in the evacuated fluid contents. We discuss the differential diagnosis and the diagnostic algorhythm in cystic cervical lesions.*

Key words: *cervical cyst, hyperparathyroidism, diagnosis*

Corresponding author: *Dr. Yordan Vlahov, MD, MBA, Clinic of Endocrinology and Metabolic Diseases, University Hospital Sofiamed, Sofia, Bulgaria, e-mail: jordanvlahov@gmail.com*

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INTRODUCTION

Primary hyperparathyroidism (PHPT) is the third most common endocrine disorder after diabetes and osteoporosis [1]. PHPT is a calcium-phosphate metabolism abnormality in which excessive parathyroid hormone (PTH) secretion by one or more parathyroid glands (normally four) leads to hypercalcemia [2].

The incidence of PHPT is approximately 66/100 000/year (between 34 and 120/100 000/year) in women and 25/100 000/year (between 13 and 36/100 000/year) in men. The incidence increases with age, with the increase being more pronounced in women – in the age group below 50 years the prevalence is 80/100 000/year in females and 36/100 000/year in males, and for the age group 70-79 years – 196/100 000/year and 95/100 000/year, respectively [3-7]. The prevalence and incidence are higher in countries with regular biochemical screening and where PHPT is actively sought.

In approximately 75-85% of the cases PHPT is due to solitary benign parathyroid adenoma, in 2-12% – two adenomas, and in 1-2% three of the parathyroid glands are affected by adenomas, rarely all four glands are involved (mainly in patients with multiple endocrine neoplasia [MEN] – MEN1, MEN2a, MEX2b, or hyperparathyroidism with tumor of the jaw [HP-J syndrome]), and in approximately 1% of the cases parathyroid cancer is present [8]. PHPT can be asymptomatic or affect the bones (osteoporosis, bone cysts and fractures), the kidneys (nephrolithiasis/nephrocalcinosis), or the gastro-intestinal tract (the first descriptions of the disease in 1930s from Massachusetts General Hospital report peptic ulcers in 8% of the patients with PHPT [9]).

The main criteria for the diagnosis are hypercalcemia and increased PTH inadequate to the high calcium. In countries with pro-active screening the prevalence of normocalcemic patients is higher. If early detected and treated/operated, the prognosis is favorable. The formation, secreting PTH, can be visualized using ultrasound examination of the neck which reveals a well-defined and well vascularized solid hypoechogenic lesion behind and/or beneath one of the thyroid lobes. The precise localization of the lesion is made using 99mTc MIBI scintigraphy, or more recently – using SPECT-CT.

We present a patient with PHPT that manifested with a large cystic formation on the neck.

CASE PRESENTATION

A 73-years-old Caucasian female patient was admitted to the Clinic of Endocrinology and Metabolic Bone Diseases for diagnostic evaluation of a lump in the left anterior cervical area that appeared about a year before the admission and lead to hoarseness, tightness sensation in the neck and difficulties in swallowing (dysphagia). The patient reported that in 2003 she underwent resection of the thyroid for nodular goiter (struma nodosa). The ultrasound examination revealed a cyst in the left neck region. Due to symptoms of compression the patient was offered surgical treatment but she refused and was referred for cyst puncture and sclerosing with absolute alcohol.

At the admission soft elastic formation on the neck, engaging the left neck region at the lower pole of the left thyroid lobe was present. The formation was painless and protuberated above the neck (Fig. 1).

The ultrasound examination revealed a large cyst at the lower pole of the left thyroid lobe 35 x 33 x over 35 mm that dislocated the trachea to the right (Fig. 2).



Fig. 1. Formation in the left cervical region

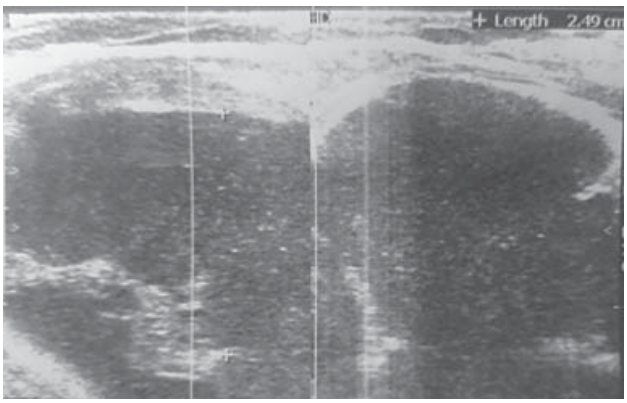


Fig. 2. Ultrasound examination of the neck before the cyst puncture – a cystic formation with fluid content

The cyst was punctured and 30 ml bloody fluid were evacuated. That lead to decrease of cyst volume to approximately 90 cm^3, but compression of the trachea with deviation to the right persisted (Figure 3).

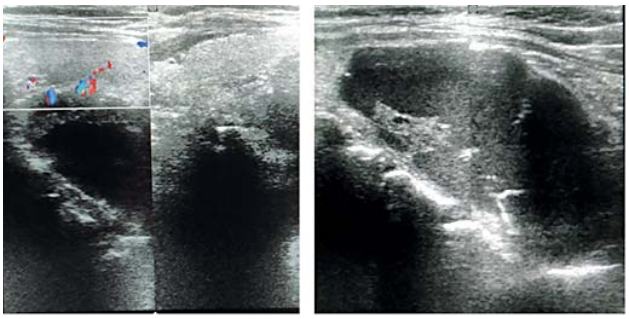


Fig. 3. Ultrasound examination after the puncture – decreased cyst volume with persisting deviation of the trachea

The localization of the formation behind and beneath the left thyroid lobe suggested the possibility of parathyroid origin of the cyst and calcium-phosphate metabolism investigations were performed. The clinical-laboratory constellation of PHPT was detected: hypercalcemia (3.52 mmol/l; /2.2-2.65/); high PTH (1030.06 pg/ml; /12-88/); hypophosphatemia (0.73 mmol/l; /0.81-1.45/); preserved renal function CKD-Epi = 60 ml/min. The levels of PTH inside the punctuate reached very high levels (above 3 507 pg/ml [12-88]). These investigations confirmed the parathyroid origin of the lesion. SPECT-CT was performed in order to rule out the presence of additional parathyroid lesions. The SPECT-CT revealed a solitary mixed type formation in the left neck region that was partially situated behind the clavicle and dislocated the trachea to the right (Fig. 4 and 5).

The patient was started on cholecalciferol for prophylaxis of recalcification tetany and was referred for surgical treatment. During the surgery, a capsulated

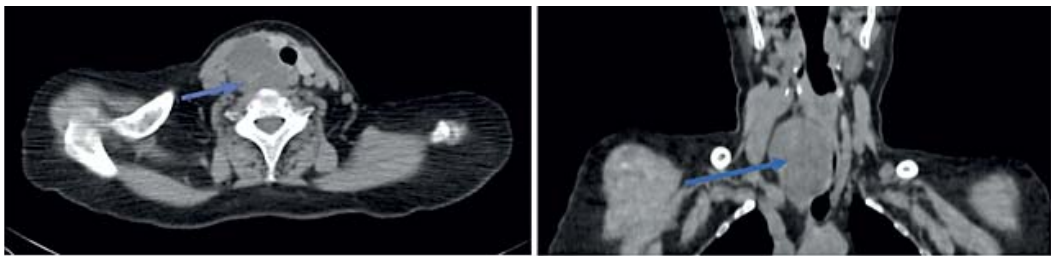


Fig. 4. Computed tomography (CT) of the formation (arrow)

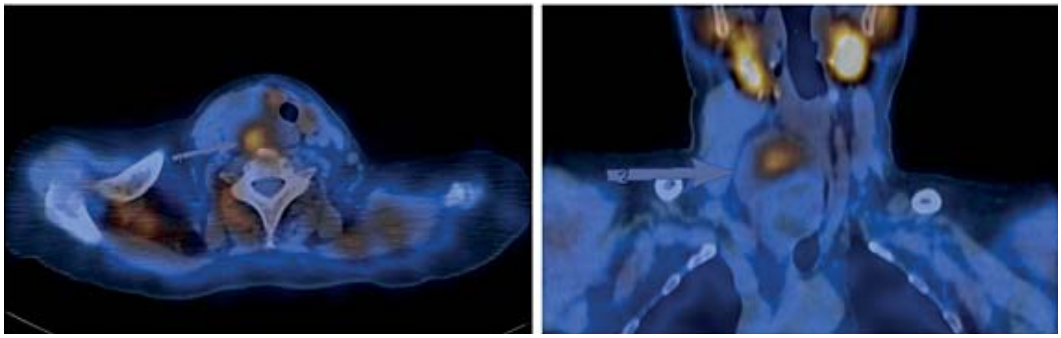


Fig. 5. SPECT-CT of the neck – solitary mixed type formation in the left neck region, partially situated behind the clavicle, dislocating the trachea to the right

cystic formation 75 x 65 x 45 mm filled with brownish fluid was extirpated. The histological examination revealed cystic parathyroid adenoma covered mainly with monomorphous chief parathyroid cells.

During the early postoperative period serum calcium reached normal levels and the patient developed no symptoms of tetany.

DISCUSSION

PHPT is characterized by excessive secretion of parathyroid hormone (PTH), a polypeptide hormone consisting of eighty-four amino acids, that leads to hypercalcemia. It is also defined as increased PTH that does not correspond to the increased calcium plasma levels, or as unregulated overproduction of PTH leading to abnormal calcium homeostasis. PHPT can be asymptomatic for a long time and after apoplexy of the parathyroid adenoma large cystic formations compressing the adjacent structures can develop. In our patient a residual cystic formation due to apoplexy of clinically silent parathyroid adenoma has lead to the diagnosis of PHPT. The localization of the lesion – behind and beneath the left thyroid lobe, further suggested parathyroid origin of the lesion and directed us towards evaluation of calcium-phosphate metabolism. Non-secreting parathyroid cysts are rare and in such cases calcium, phosphate and PTH blood levels are within the normal limits. The cyst puncture usually shows clear fluid with high PTH content.

As first and most probable diagnosis, the patient's age and gender suggest hemorrhage in thyroid formation, because the prevalence of thyroid diseases in Bulgaria is high. The prevalence of nodular goiter in the Bulgarian population is 24.4% (32.1% in females and 15.7% in males), increasing to above 50% in females > 70 years of age [10]. Therefore, thyroid gland formation was the initial diagnosis at the admission and sclerosing of the cyst was planned because the patient refused surgical treatment. PHPT was a far less probable diagnosis, because the prevalence of hyperparathyroidism in Bulgaria is relatively low – 3.59% of the population, with marked increase from 1.9% in young adults to 6.8% of the population above 60 years of age (in 8.3% of the women and 4.7% of the men in this age group) [11]. Therefore, PHPT is a rare differential diagnosis in patients with cysts in the neck region.

This clinical case suggests that all patients with formations in the neck, especially women above 70 years, should be investigated for calcium-phosphate metabolism and are indicated for specific imaging and hormonal studies, and should be referred to endocrinology clinics for evaluation.

Disclosure Summary: *The authors have nothing to disclose.*

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