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Case Report

Cancer of ectopic parathyroid gland presentation of the disease with a case report

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Abstract: Tc-99m-methoxyisobutylisonitril (MIBI) scintigraphy is localizing diagnostic methods that is used for detection of sicken parathyroid gland (PT). The use of this method for PT diseases diagnosis makes surgical treatment of a patient more successful. This is a report about the patient who was surgically treated for primary hyperparathyroidism caused by hyperplasia of parathyroid glands and cancer of ectopic parathyroid gland. He was operated in two acts. The first surgical intervention was performed without preoperative diagnostics of Tc-99m-MIBI scintigraphy, while the second surgical intervention was preceded by Tc-99m-MIBI scintigraphy which clearly showed the existence of tumor in the back mediastinum (ectopic parathyroid gland). Pathophysiological analysis of the extirpated parathyroid gland showed the case of ectopic parathyroid gland cancer. The use of Tc-99m-MIBI scintigraphy as a localizing method before the first surgical intervention could have saved the patient from the second one and from risks it could have caused, as well.

Keywords: Tc-99m-MIBI scintigraphy, primary hyperparathyroidism, cancer

Introduction

Primary hyperparathyroidism is a disease characterized by increased secretion of parathyroid hormone (PTH) caused by hyperfunction of one or more parathyroid glands. Enlarged PTH concentration in the circulation causes hypercalcemia and hypophosphatemia. Clinical manifestation of the disease can be hardly noticeable and it can have a benign course for many years. The disease can sometimes start suddenly with life-threatening complications (dehydration and coma), known as hypercalcemic parathyroid crisis. Those who have PHPT can have numerous symptoms and signs, such as kidney calculosis, peptic ulcus, constipations, mental changes, cardiovascular disorders, and demineralization of bone tissue in more serious cases of PHPT. Direct cause of PHPT genesis is unknown but it is considered to be genetically conditioned [1-3]. Hyperplasia, adenoma, and PT cancer are basic PHPT pathoanatomical substrates. In hereditary cases demonstration is either isolated – familial hyperparathyroidism (in which case only primary hyperparathyroidism is hereditary) or integral part of multiple endocrine neoplasia type 1 and 2 (MEN1 and MEN2). X-ray of head and neck area is considered as one of the etiologic factors of sporadic hyperparathyroidism. Cancer as a cause of PHPT is rare and it has been found in 0.5–5% cases of PHPT [1-3]. PHPT diagnosis is strictly established by laboratory analyses [4]. Basic parameters are: hypercalcemia, enlarged parathyroid hormone, hypophosphatemia, and calciuria. Symptomatic PHPT treatment is strictly surgical. Failure of surgical PHPT treatment is about 10% and this can be imputed to inadequate preoperative localization of PHPT causes [5]. Localizing examinations can be non-invasive and invasive. Non-invasive methods include ultrasound diagnostics (U), scintigraphy (Tc-99m-MIBI scintigraphy is mainly used), computerized tomography (CT), magnetic resonance (MRI), and X-ray diagnostics [6]. Biopsy with a thin needle under the control of ultra-

Table 1. Laboratory analyses before and after the operation

Laboratory parameter	Preoperative	Postoperative
Serum calcium	3.41	1.93
Inorganic phosphate	0.50	1.11
Magnesium	0.35	0.71
PTH	1,270.00	15.60

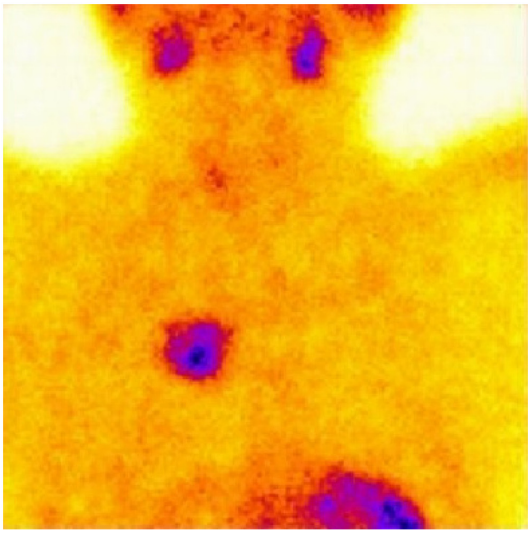

Figure 1. Late phase of parathyroid gland scintigraphy. Isotope Tc-99m-MIBI.

sound, selective arteriography, and selective vein catheterization are used in invasive localizing examinations.

Case description

A 54-year-old male patient had many difficulties for a few last years: renal colics, polyuria, depression, pains in his muscles and bones, hypertension, as well as pathologic fractures of left lower leg and left forearm. After several hospitalizations, a primary hyperparathyroidism was diagnosed; diagnosis was based on laboratory findings (serum calcium, PTH). It was determined by standard diagnostic procedures (US, scintigraphy, hormone status) that there was inactive solitary nodus in the left lobe of thyroid gland. Enlarged values of serum calcium, low values of inorganic phosphates, and high values of PTH (**Table 1**) were laboratory confirmed. After the preoperative preparation which consisted of implementation of calcium antagonists (30 mg of pamidronate-Aredia), bisphosphonates, corticosteroids (last value of serum calcium was 2.86 mmol/L), and infusions of crystalloid solutions, the patient was operated on. Left lobo isthmectomy, right subtotal parathyroidectomy, and left parathyroidectomy with resection of cervical thymus were performed. PH finding showed hyperplasia of removed parathyroid glands. PH finding: Primary, nodular hyperplasia of PT with dominance of main cells and scattered follicular histological organization. Soon after the surgery, a slight fall of serum calcium value (2.87 mmol/L) occurred. The patient was released. Symptoms of PHPT were still present at the checkup. Laboratory analyses showed enlarged values of serum calcium (3.16 mmol/L), low values of inorganic phosphate (0.36 mmol/L), and enlarged values of PTH (1150 pg/mL), which indicated the existence of ectopic parathyroid tissue. To localize it, we used Tc-99m-MIBI scintigraphy, and computerized tomography (CT). Tc-99m–MIBI scintigraphy registered pathologic radio-markers accumulation in right upper half of the back mediastinum. Diphasic scintigraphy of neck and mediastinum area was performed 10 minutes after the initiation of 555MBq Tc-99m-MIBI, and again 2 hours later. Previously made scintigrams (thyroid phase) showed radio-drugs accumulation in projection of upper mediastinum (close to lung apexes). The second scintigram (parathyroid phase), after it was rinsed out, showed a slight radio-drugs accumulation in projection of the thyroid gland remainders; the area of radio-drugs was also detected in the upper mediastinum (along the lung apexes, mostly on the right side), which scintigraphy matches to ectopically localized tissue of parathyroid gland (**Figure 1**). Computerized tomography with the window for the mediastinum in the middle back mediastinum retro and in right parallel indicated a multilocular septal tumor shade with peripheral calcificate and septa calcification, attenuation of soft tissues sized 45*55*65 mm. The change dislocated esophagus and trachea to the opposite side and it was in a close contact with ascending aorta. The second surgery consisted of partial sternotomy and extirpation of the tumorous change sized about 6*10 cm. PH finding of the extirpated tumor change: Carcinoma glandulae parathyroideae (middle differentiated histologic type) (**Figure 2**). Postoperative course went on without any surgical complications. The value of serum calcium and parathormone fell down soon after the

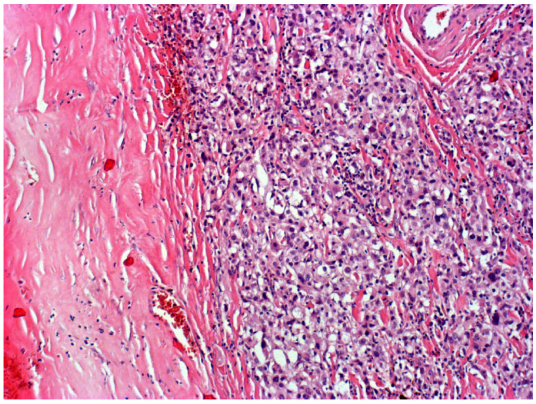

Figure 2. Carcinoma glandulae parathyroideae (middle differentiated histologic type).

surgery (**Table 1**). Postoperative substitutional therapy (Ca-Sandoz, $CaCl_2$, Alfacalcidol, Mg) was implemented. Six days after the surgery the patient was transferred to Metabolic Department of Internal Clinic where his treatment was continued with calcium medicines, magnesium, active type of vitamin D, ACE inhibitors, calcium antagonists, antibiotics, low-molecular heparin, followed by constant monitoring of electrolytic, acid-basic and chemodynamics status and necessary correction of them. Period of parenteral compensation lasted for a long time (about 3 months) which explains the intensity and duration of PHPT. Implemented therapy led to subjective and objective improvement so recovered and examined patient was released to further ambulatory treatment with following therapy: Calcium gluconate, ascorbic acid 500 milligrams tablets 2+2+2, Alfacalcidol 0.25 milligrams tablets 2+2+2, Magnesium hydroxide 150 milligrams tablets 1+0+1, Quinapril 20 milligrams tablets 1+0+1, Amlodipine 5 milligrams tablet 1+0+0.

Discussion

Laboratory diagnostic methods (values of serum calcium, calciuria, and PTH) are the basis for establishing the diagnosis of parathyroid glands diseases [1-4]. Surgical treatment of parathyroid gland diseases demands their precise localization which emphasizes the importance of localizing examinations in preoperative procedure [5-7]. Localizing examinations can be invasive and non-invasive. Non-invasive methods are: ultrasound diagnostics (US), scintigraphy (Tc-99m-MIBI scintigraphy has priority), computerized tomography (CT), and magnetic resonance (MRI). Biopsy with a thin needle under the control of ultrasound, selective arteriography, and selective vein catheterization are used in invasive localizing examinations.

Ferlin was the first who used double radio-isotopic scintigraphy with 201Tl-chloride and Tc-99m pertechnetate in order to visualize parathyroid glands [8]. There are many controversies about the usage of localizing examinations before the first operation. Preoperative localizing examinations are not used for the first operation in most of the cases mostly because of the general opinion that an experienced surgeon will almost always find sickened parathyroid gland during the first operation. This attitude is supported by the fact that localizing examinations are of little importance for hyperplasias of parathyroid glands. Doppman, the radiologist, has the same opinion; he thinks that the only thing important for the localization of the pathologically changed parathyroid gland is an experienced surgeon [9]. However, frequent need for the second operation in PHPT treatment has imposed the need for localizing examinations [10-12].

Conclusion

Localizing methods, especially Tc-99m-MIBI scintigraphy, CT, MRI, and US are important in preoperative diagnostics of pathologic conditions of parathyroid gland. The presented case shows that implemented Tc-99m-MIBI scintigraphy in combination with CT localized etiologic cause of PHPT; it also shows that the usage of localizing diagnostic procedures would reduce the need for secondary surgery and all the risks that might appear.

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