



## A Rare Presentation: Parathyromatosis

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### Abstract

Parathyromatosis is a rare condition which occurs after parathyroidectomy and it is a situation that is difficult to diagnose and treat in hyperparathyroidism. Parathyromatosis would result from seeding after the fracture of the parathyroid gland capsule during parathyroid surgery as well as there are some other etiological factors either. We here aimed to mention about etiology, diagnosis and treatment options of parathyromatosis with presented our case.

### Introduction

Parathyromatosis is a rare cause of hyperparathyroidism. When we look at literature; two causes were defined for its occurrence. In primary type, it might be due to hyperplasia of the parathyroid residues and on the other hand in secondary type generally it might be due to planting after parathyroid surgery. In secondary type residual thyroid tissues stimulate due to existing chronic renal failure. In some studies as a third theory; it was said that it is a low grade malignancy [1,2]. In this case; we aimed to present a parathyromatosis case with surgery history due to parathyroid adenoma and who had hypercalcemia in examinations which were conducted prior to the pilonidal sinus surgery.

### Case Presentation

Thirty two year old male patient applied to our hospital general surgery polyclinic for pilonidal sinus surgery. In his history; there was only weakness complaint and discharge from presacral region. It was learned that he had surgery with the diagnosis of parathyroid adenoma and pathology results were parathyroid adenoma. As his routinely performed preoperative laboratory test result was Ca<sup>++</sup>: 13.2 mg/dl [8.4-10.2]; parathyroid hormone levels were checked. Parathyroid hormone was high as 338 pg/ml [15-65]. Kidney function tests were normal. In his neck ultrasonography; about 24 mm × 10 mm × 13 mm sized solid lesion (parathyroid adenoma? carcinoma?) was detected which was localized beyond thyroid borders at inferior of right thyroid lobe and in Doppler ultrasonography it was hypervascular, with lobule contours and heterogeneous hypoechoic structure. In his parathyroid scintigraphy; pathological activity retention was described in favor of parathyroid adenoma at inferior part of right thyroid lobe (Figure 1). Urinary tract ultrasound and bone densitometry were found to be normal. In computed tomography of the neck and mediastinum; 11 mm × 10 mm sized hypodense lesion at posteroinferior of right thyroid lobe was reported. In September 2014, the patient was taken to surgery with these findings and as a result of 1cm sized adenoma at lower right are of parathyroid and micronodular view around it in the surgery; unilateral thyroidectomy (right), right lower parathyroidectomy, and bilateral central neck dissection were made with possible diagnosis of parathyroid carcinoma. Control parathyroid hormone after surgery was 7.94 pg/ml [15-65]. Pathological result was Parathyromatosis (Figure 2).

### Discussion

Parathyromatosis is a rare condition which occurs after parathyroidectomy and it is a situation that is difficult to diagnose and treat in hyperparathyroidism. After being described by Palmer for the first time in 1975, about 35 cases have been reported in the literature [3]. Although parathyroid residues can be seen in thyroid gland and cervical lipid tissue; they generally located in thymus in parathyromatosis cases due to embryological residues. Although Parathyromatosis is a benign disease with slow growing rate in the area that it's located at; you should be protected from effects of long term exposure to hyperparathyroidism [4].

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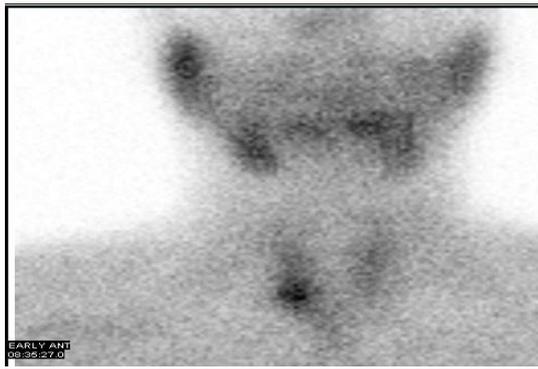


Figure 1: Patients MIBI scan.

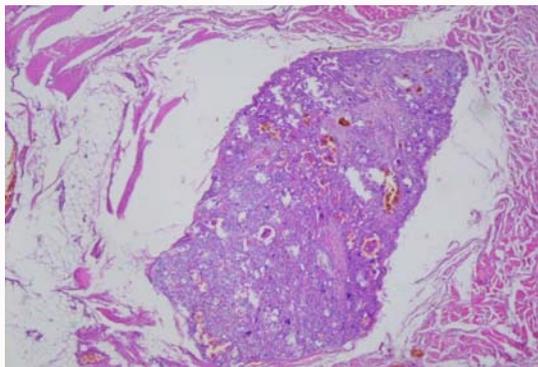


Figure 2: Striated muscle tissue and fat tissue nodules in parathyroid tissue (H&Ex40).

In necessary cases; Parathyroid can be fed by diffusion when they are planted to the muscle or fat tissue, can make growth by angiogenesis and they can survive at where they are [4]. Rupture of capsule during parathyroidectomy, can lead to dissolution of tissue and proliferation of that tissue in time. In this sense, surgeon can be effective in etiology. In case of renal failure, this spilled parathyroid's can be hyperplasia under stimulation of secondary hyperparathyroidism. If capsular rupture happens during surgery, parathyromatosis can happen at a certain point during surgical plan. When we look at the literature; parathyromatosis occurrence after parathyroid surgery is defined between a time intervals of 5 months to 19 years [5]. In 22 of 35 patients with Parathyromatosis, chronic renal failure was detected [6].

Parathyromatosis mostly occurs in women and in the fifth decade and in studies the difficulty for diagnosis is mentioned. As a cause of this difficulty; being a diagnosis which is usually unthinkable is specified. Only 4 cases were reported in literature which had diagnosis before the surgery [7]. Hyperparathyroidism can cause kidney stones, bone pain, gastrointestinal and psychiatric symptoms. In laboratory tests; blood calcium levels can be high (1-2 mg/dl and above), in patients with chronic renal failure normal calcium levels can be found. In imaging methods which were used before neck exploration; less than 50% of the patients can have diagnosis, thus ultrasonography is one of the most helpful method in diagnosis. In the differential diagnosis of Parathyromatosis; colored Doppler ultrasonography can be helpful for distinction of other cervical malignancies and gray scale ultrasonography may show similar characteristics. MIBI SPECT, can be usually used for localization of hyperactive parathyroid tissue [8].

In differential diagnosis; parathyroid adenoma and carcinoma

should be considered. In all of these clinical conditions; there are persistent and high levels of parathyroid hormone. In patients with parathyroid carcinoma, calcium levels are markedly higher. Definitive diagnosis is established histologically. There are studies in literature about sowing while making fine needle aspiration biopsy ultrasonographically with preliminary diagnosis of parathyroid adenoma in order to make histopathological diagnosis or look for parathyroid hormone but currently there is no proven information [9,10]. Macroscopically; it can be soft, whitish yellow, in size ranging from microscopic size to 2 cm. Adenoma and carcinoma can be observed as a single structure and on the other hand Parathyromatosis can be observed in a multi glandular form. Histological difference of Parathyromatosis from cancer or adenoma is the lack of real capsule of the parathyroid tissue [11]. In carcinoma there can be invasion to adjacent tissues, lymph node or distant organ metastasis. In Immunohistochemical studies, we can use some markers (retinoblastoma expression, parafibromine loss, Galectin-3 over expression) for the differential diagnosis of parathyroid carcinoma [12].

While it is difficult, the treatment of Parathyromatosis surgery. All of the possible foci can be removed with an ultrasonography which is used during surgery. However, if differential diagnosis cannot be made during surgery, it is suggested to behave as a carcinoma and make the surgery. Medically, calcium and parathyroid hormone levels can be decreased by using calcimimetic drugs (Cinacalcet) and bisphosphonates [13,14].

In conclusion; Parathyromatosis, is a rare cause of hyperparathyroidism. It should be considered in patients with previous parathyroid surgery, high levels of calcium and PTH. For diagnosis; Histopathologic examination is necessary and in treatment first surgical procedure should be made carefully and if parathyromatosis is developed, surgery should be made with extensive exploration. Medical treatment can be tried if surgery is not effective or there is a contraindication for surgery.

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