

Letter to
Editor**A Rare Case of Multiple and Ectopic Parathyroid Adenoma**Author(s)  Fatih Güzel, Şadiye Altun Tuzcu

Affiliation(s) Dicle University Faculty of Medicine, Department of Nuclear Medicine, Diyarbakır, Turkey

Corresponding Author Şadiye Altun Tuzcu, Dicle University Faculty of Medicine, Department of Nuclear Medicine, Diyarbakır, Turkey
E-mail: sadiyetuzcu@yahoo.com.tr

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JEIMP belongs to "The Foundation for the Management of Chronic Diseases" and is supervised by the MKD Digital Publishing. www.jeimp.com and digitalmkd.com**Dear Editors,**

Primary hyperparathyroidism (P-HPT) is an endocrine disorder caused by the autonomous overproduction and secretion of parathyroid hormone (PTH) by one or more abnormal parathyroid glands. Patients are mostly between the ages of 40-70, with a higher incidence in females compared to males. Most patients with P-HPT have hypercalcemia and elevated PTH levels (5,6). In primary HPT, the main issue is the growth of one or more abnormal parathyroid glands and the inappropriate secretion of PTH from these glands relative to serum calcium levels. Solitary parathyroid adenoma and diffuse parathyroid hyperplasia, more rarely multiple adenomas, and parathyroid carcinoma, are significant pathologies resulting in primary hyperparathyroidism. Pathologically functioning parathyroid cells in parathyroid adenoma show lower sensitivity to high calcium levels than normal. Therefore, since the serum calcium set point is at a higher threshold, the circulating calcium level is maintained at much higher levels. In parathyroid hyperplasia, there is a general increase in the number of parathyroid cells that produce and secrete excess PTH but maintain normal sensitivity to calcium. Both conditions cause hypercalcemia, but there is a difference in PTH levels (7). In P-HPT cases; solitary parathyroid adenoma is the most common cause, accounting for 85-90%. Various literatures report multiple parathyroid adenomas at different rates, ranging from 2-11%. Other causes of primary hyperparathyroidism include parathyroid hyperplasia, which accounts for 15% of all cases, while carcinomas, which are rarer, are responsible for only 1% of cases (1,2). In the treatment of P-HPT; the traditional surgical approach is the removal of all hyperfunctioning parathyroid glands. This is possible with a good parathyroidectomy operation that includes bilateral neck exploration. Bilateral neck exploration is based on the evaluation of all parathyroid glands and then the removal of one or more glands that appear pathological (5). However, due to the long duration

of the operation and the high rates of mortality and morbidity, preoperative diagnostic studies are becoming increasingly important. The diagnosis of P-HPT is made with clinical and laboratory examinations. The purpose of parathyroid imaging is to locate abnormal parathyroid glands in the preoperative period, to shorten the operation time, and to reduce morbidity rates (8). In this context, parathyroid scintigraphy with technetium-99m-hexakis-2-methoxy-isobutyl isonitrile (Tc-99m MIBI) is a current method widely used to determine the presence, number, and localization of parathyroid adenomas. In our case, we aimed to provide an overview of the imaging and treatment processes through a case of four parathyroid adenomas, one of which is ectopically located, a rare condition detected by Tc-99m MIBI parathyroid scintigraphy.

Case

A 21-year-old female patient presented with widespread bone pain. Investigations revealed high serum calcium and parathyroid hormone levels, and bone densitometry showed low bone mineral density for her age. A diagnosis of primary hyperparathyroidism was made. Preoperative ultrasound examination detected suspicious nodules compatible with parathyroid adenomas: two in the right lobe and one in the left lobe of the thyroid gland. Additionally, preoperative Tc-99m MIBI parathyroid scintigraphy reported suspicious activity consistent with adenomas in both lobes, persisting in early (20 min) and late images (120 min) (**Image 1**). During surgery, three parathyroid glands were excised with the support of an intraoperative gamma probe, and adenomas were confirmed in frozen sections. However, due to the lack of significant improvement in intraoperative parathyroid hormone levels, a complementary thyroidectomy was performed. Subsequently, a parathyroid adenoma located within the thyroid tissue was identified in the thyroidectomy specimen. Postoperative serum calcium and PTH levels showed significant improvement.

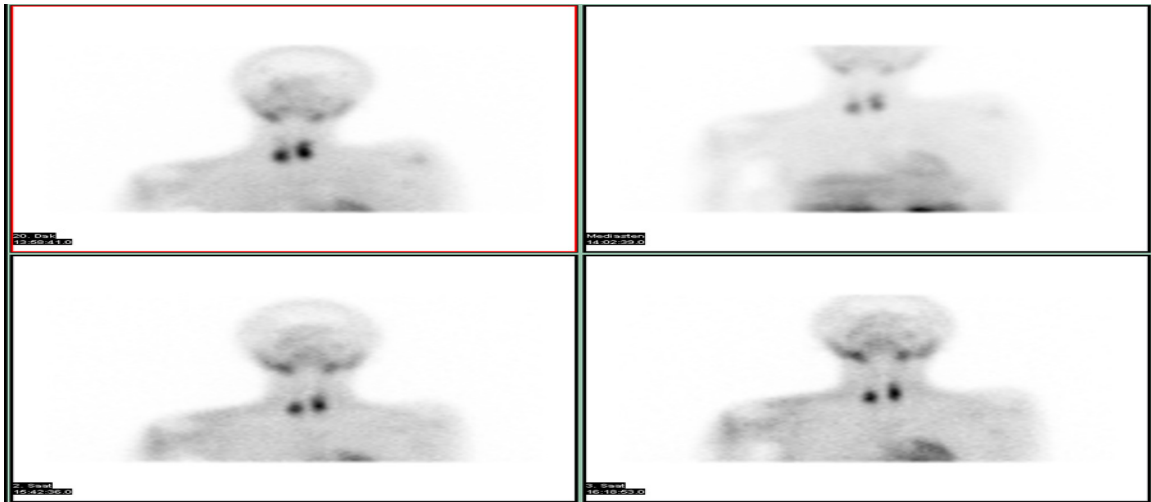


Image 1. Tc-99m – MIBI Scintigraphy: Persistent activity in early and late images in the thyroid gland region.

Multiple parathyroid adenomas have been reported in 2-11% of cases with P-HPT (2). In 6-16% of cases, one or more hyperparathyroidism adenomas are found in an ectopic position. Multiple and ectopically located parathyroid adenomas, though rare, are encountered in routine endocrine practice and often present with persistent and recurrent hyperparathyroidism (3,4). 80-95% of patients with P-HPT are treatable with a simple parathyroidectomy after the first surgery (9). In cases where a cure is not achieved, persistent hypercalcemia immediately after surgery or recurrent hypercalcemia after a long period of normal serum calcium levels is observed (10). In our case, despite the significant contribution of preoperative Tc-99m MIBI scintigraphy in identifying multiple parathyroid adenomas, the persistent intraoperative hyperparathyroidism necessitated the search for a new ectopic adenoma focus. With the help of intraoperative gamma probe signals from thyroid tissue, an ectopically located intrathyroidal adenoma was detected. We confirmed that essential components of curative treatment in parathyroid adenoma cases are preoperative scintigraphic imaging (USG and Tc-99m MIBI scintigraphy) and intraoperative PTH measurements. In this context, it should not be forgotten that in primary hyperparathyroidism cases, Tc-99m-MIBI scintigraphy and intraoperative gamma probe use are crucial for localizing hyperfunctioning parathyroid pathologies preoperatively and especially for identifying hyperfunctioning adenomas with typical or atypical locations in cases of resistant hyperparathyroidism in parathyroid surgery (2).

In conclusion, Tc-99m MIBI parathyroid scintigraphy, widely used for determining the number and location of lesions before adenoma surgery in patients newly diagnosed with primary hyperparathyroidism, is a current method that reduces the duration, complications, and scope of the procedure.

DECLARATIONS

Conflict of Interest Statement: The author declares no conflicts of interest related to this letter.

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